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Content

Lean and Seamless

In rehabilitation, an ultimate goal is to restore functions of an individual so that he/she reaches independency in performing activities in daily life and has better quality of life. To achieve the goal, rehabilitation physicians (physiatrists) as well as other rehabilitation professionals should learn and apply business knowledge and skills to improve rehabilitation services so that those who need rehabilitation could access to the services and truly get benefits.

In business, a lean process is a method of eliminating wasteful practices to improve service efficiency and effectiveness. The concept of lean has been implemented in healthcare services including rehabilitation as rehabilitation services delivered by rehabilitation team of multi-professionals are complex. Many processes may be unnecessary and should be removed. All professionals involved have to work together to set common goals and to produce a coordinated set of therapy.

Among rehabilitation professionals, nurses and occupational therapists both work to improve patients' performance in self-care. Each discipline has one's own principles of approach. Breaking down boundaries and complexity would lead to a seamless flow of services, and thus benefit the patients. The concept of seamless patient care consists of consistency, continuity and coordination of care that should be implemented across the whole work processes. Self-care by nurses and self-care training by occupational therapists may vary and confuse patients. Cross functional communication is necessary to provide well-coordinated, consistent and continuous services across disciplines. Moreover, all in the team should work together to clarify responsibilities, care objectives, and treatment plans.

In this issue, there is an interesting research study reporting a successful seamless care and training in self-care between nurses and occupational therapists. The seamless self-care training firstly delivered by occupational therapists is continued consistently and efficiently by nurses, and efficiently makes patients with stroke effectively achieve independency in dressing within a short period of time. This seamless care may be an appropriate solution for hospitals where a number of occupational therapists is limited. Lastly, just reading the article makes readers learn and know more but a true success is to apply it in real practice.

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Apichana Kovindha, MD, FRCPhysiatrT
Editor, ASEAN J Rehabil Med

Original Article

The Outcomes of Seamless Self-care Training between Occupational Therapist and Nurse in Stroke Patients

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ABSTRACT

Objectives: To investigate the outcomes of seamless self-care training to stroke patients by occupational therapists (OTs) and nurses.

Study design: Quasi-experimental research design.

Setting: Rehabilitation ward, Siriraj Hospital, Mahidol University.

Subjects: Thirty-five inpatients with first stroke, normal mental functions and age between 18 to 75 years old, were included in the study. All needed help in dressing and washing oneself activities.

Methods: Participants were divided into two groups. The conventional group received self-care (dressing and washing oneself) training from OTs for 3 days; on day 4, OTs supervised them, thereafter nursing staffs were responsible for such activities in a traditional nursing care until day 14. The experimental group received a "seamless self-care training", in which OTs trained patients for 2 days, supervised them for the next 2 days, and the nursing staffs joined the OTs on day 4 to learn how to facilitate the patients and led the training until day 14. The primary outcome was a normalized gain of the self-care score. Secondary outcomes were a modified Barthel index (MBI) score, self-efficacy, and patient satisfaction. The outcomes were evaluated at baseline (day 1) and after completing the training (day 14).

Results: After adjusting by normalized gain calculation, there were significantly more participants in the experimental group who achieved a higher gain level (0.7-1) than in the conventional group (p = 0.028). The experimental group had considerably higher self-efficacy for washing oneself activity after the training (p = 0.013) and were more satisfied with the training program than the conventional group. The MBI improved in both groups.

Conclusion: This seamless self-care training with good collaboration between occupational therapists and nursing staffs during inpatient rehabilitation was proved to be effective in dressing and washing oneself training and promoting self-efficacy of stroke patients

Keywords: self-care, nursing care, occupational therapy, inpatient, stroke rehabilitation

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Introduction

Stroke is a leading cause of death and disabilities worldwide. Estimates from the Global Burden of Disease Study in 2017 ranked stroke as the third most common cause of disability-adjusted life-years (DALYs).[1] Despite the development of stroke care, many stroke survivors are left with functional deficits.[2] Activities of daily living (ADLs) are essential functions that all survivors need to regain their ability to live independently and their self-esteem, and to alleviate the burden on caregivers. The loss of independency with not being able to perform ADLs contributes to institutionalization and reduces quality of life.[3]

Rehabilitation is mandatory to promote functional recovery and minimize the distress of stroke survivors as much as possible. Even though rehabilitation outcomes are expected in all aspects, in one study the highest improvements were observed for bowel and bladder functions, transfer, and mobility, while the lowest improvements were seen in bathing (washing oneself), grooming (caring of body parts), dressing, and stair climbing.[4]

Occupational therapists (OTs) are responsible for assessing and teaching skills to facilitate patients' activities and participation, including maximizing their performance of ADLs, and provide training programs and techniques that are tailor-made for each individual's impairment using targeted task-specific training, which has been acknowledged as the most effective method for enhancing functional outcomes.[5-7] Nevertheless, the most important key factor for successful training outcomes is the collaboration of multiple healthcare professionals. All team members, especially nurses who spend lots of time with patients, should focus on encouraging and facilitating patients in transferring the newly relearned skills from professionals into their lives. Unfortunately, each expert usually approaches the patients from their own aspects, causing a lack of team perspective that could potentially preclude the patients from achieving their rehabilitation goals.[6,8]

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In current inpatient rehabilitation practice, there is still a gap in carrying out self-care training. Self-care training is usually carried out by OTs at the occupational therapy unit, whereas nursing staff assists washing and dressing the patients during admission at the rehabilitation ward. Due to time constraints, nurses usually concentrate on preparing the patients to be ready on time for other specific training.[9] This hinders them from allowing patients the time and opportunity to practice by themselves, resulting in patients' having low confidence and a perception of low self-efficacy. Consequently, some patients may not develop sufficient skills to perform self-care independently after discharge. This situation was reported by Pryor and Long that an integrative function of nurses and other professionals was sub-optimal.[9,10]

Therefore, in the present study, we aimed to develop and implement an interdisciplinary team model with integration of all the relevant expertise and to establish powerful collaboration and communication among the team. To this extent, we designed a new approach toward self-care training called the "seamless self-care training" technique, which involves enhanced coordination and communication between OTs and nurses in the rehabilitation ward to promote interdisciplinary cooperation in self-care training. We hope that this technique could make therapeutic intervention more successful and fill the gap in treatment processes, as well as to increase the self-efficacies of stroke survivors. The objective of this study was to explore the outcomes of providing this seamless self-care training to stroke patients.

Methods

This quasi-experimental research was conducted in an inpatient rehabilitation setting at the Department of Rehabilitation Medicine, Faculty of Medicine Siriraj Hospital, Mahidol University, Thailand. All patients included in this study or their legal guardians provided a written informed consent. The Siriraj Institutional Review Board approved the study protocol (SIRB COA no. Si 644/2014), and this study was registered with Clinicaltrail.gov (NCT02361307).

Participants

First-time stroke patients aged 18-75 years old who were able to sit without support and who had never received self-care training were eligible for inclusion in the study. Those who had bilateral weakness, dependent basic self-care support before the occurrence of stroke, cognitive impairment, neglect, apraxia, and depression, were excluded from the study. Upon admission, every patient was screened by the Thai Mental State Examination (TMSE). The cutoff score in the diagnosis of cognitive impairment is a score of less than 24 out of 30.[11] Depression was assessed by the Patient Health Questionnaire (PHQ)-9. The cutoff score to determine major depression is more than 9 out of 27.[12] After a written informed consent was obtained, patients were divided non-randomly into two groups: conventional group and experimental group.

Study protocol

The conventional group received washing oneself and dressing training from the same OT in the occupational therapy unit for 3 days. On the 4th day of training, the OT supervised the patients while performing these tasks in the rehabilitation ward. After that, a nursing staff took responsibility for these activities in a traditional way of nursing care until day 14. Duration of training session was one hour per day.

The experimental group received the new "seamless self-care training" technique, which was characterized by a collaboration between OTs and nurses. The training team consisted of two OTs with standardized training skills according to the self-care checklist for dressing and washing oneself (see appendix). The patients received training in the occupational therapy unit for 2 days and then were supervised by OTs at the rehabilitation ward for the next 2 days. On the 4th day of the training, one nursing staff came along with the OT in order to learn how to facilitate the patients according to their abilities to carry out these tasks. After that, the patients received further training from the nursing staff until day 14. Duration of training session was one hour per day. In this group, the nursing staff guided the patients to perform dressing and washing oneself according to the self-care checklist.

In both groups, each patient was in contact with and trained by only one OT and one nurse throughout the period of the study.

In order to reduce cross-contamination of the training techniques, each technique was delivered in separate periods of time. We first finished collecting data in the conventional group for one month and then conduct the seamless self-care training technique with the experimental group. During the study, patients received physical therapy or other treatments that did not affect dressing and washing oneself abilities.

Outcome measurement

For the main outcome, patients' abilities in dressing and washing oneself were evaluated by another OT on day 1 and day 14 of the program in both groups. However, since there were no specific detailed measurements for assessing such abilities after a stroke, the authors developed steps of dressing and washing oneself, so called "Self-care checklist for dressing and washing oneself" (see appendix). Based on task analysis, these two self-care activities consisted of 5 sequential activities: taking off a shirt, taking off pants, washing oneself, putting on a shirt and putting on pants. Each activity was composed of 4 steps, so there were 20 steps in total. Each step could be scored as 0, 1, or 2. A score of 2 meant the patients could perform the task independently, while 1 meant the patients could do the task with verbal cueing, and 0 meant the patients could not perform the task within 5 minutes or needed assistance to complete the task. Therefore, the maximum score was 40 for the independent performance measurements of dressing and washing oneself. This checklist had a high correlation with the Modified Barthel Index as analyzed with Spearman's correlation r = 0.764, $p < 0.001$.

Normalized gain

The raw score from the self-care checklist for dressing and washing oneself was used to calculate the normalized gain. Because stroke patients had varying scores for their initial dressing and washing oneself abilities, their possible window of improvement could be limited by the "floor to ceiling" effect. Therefore, it was considered that their improvements should be assessed by the normalized gain, which was introduced by Hake in 1998 as a rough measure of the effectiveness of a training course and has since become the standard method for reporting scores. Hake defined the normalized gain (g) as the ratio of the actual gain to the maximum possible gain,[13] as given in the equation:

Normalized gain (g) = (post-training) – (pre-training) / (maximum score) – (pre-training)

With a possible value between 0.0–1.0, the normalized gain can be divided into 3 levels: "high-g" for $g \geq 0.7$, "medium-g" for $0.3 \leq g < 0.7$, and "low-g" for $g < 0.3$.[14]

Modified Barthel Index (MBI) was used to assess the functional tasks of ADL by OTs. The score ranged from 0-20. The higher the score, the more independence the patients had in performing the ADL functions.[15]

Dressing and washing oneself time was measured from taking off a shirt until finished putting on pants. The shorter duration, the more capability one had.

Self-efficacy was rated by each patient using a numerical rating scale (NRS) in order to measure a self-perceived self-care ability level. The score ranged from 0-100. The higher the score, the more confidence one had in one own capability.

Patient satisfaction was evaluated by each patient using a numerical rating scale (NRS) for assessing one's contentment with the self-care training approach. The score ranged from 0-100. The higher the score, the more satisfied they were.

All the variables were assessed on day 1 and day 14 except dressing and washing oneself time that was assessed in the rehabilitation ward on day 7 and day 14 after the training was completed.

Sample size calculation

The minimum required sample size was calculated based on the outcomes of occupational therapy in the ward, in which 30% of stroke patients would show improved dressing and washing oneself abilities within 2 weeks after undertaking a conventional training. We estimated that the outcome of the seamless self-care training would be more patients gaining in such abilities, reaching up to 80%. Based on this estimation and allowing for a 5% error, a minimum sample size of 15 cases was calculated for each group, for 30 cases in total. For compensation to allow for a possible 20% dropout, 36 cases were studied in total.

Statistical analysis

Demographic and clinical variables were summarized using descriptive statistics. Continuous variables were described as mean with standard deviation (SD) and median with an interquartile range (IQR). Categorical variables were described by frequency and percentage. Statistical data analysis was performed using SPSS, Version 18.0, and a *p*-value < 0.05 was considered to be statistically significant. The differences in the outcome of self-care training and self-efficacy between the two groups were analyzed by independent t-test, or within group by paired t-test, while differences between groups covariate by day 1 were analyzed by analysis of covariance (ANCOVA). The gain level was analyzed by chi-square test for assessing trends.

Results

In total, 36 stroke patients participated in the study. One participant discontinued the program after the first day of training due to a medical condition, leaving 35 participants who received the allocated intervention. There were no adverse events in this trial. The participants' mean age was 58.3 (SD 10.5) years old and 21 (60%) of them were male (Table 1). Most participants (97.3%) were right handed, while 48.6% had right-side weakness. All participants needed assistance with dressing and washing oneself.

The outcome self-care scores were measured at baseline (day 1) and at day 14. After adjusting by the normalized gain calculation, significantly more participants in the experimental group achieved a higher gain level (0.7-1) than in the conventional group ($p = 0.028$). Comparing the dressing and washing oneself time taken on day 7 and day 14, both groups showed reduced time spent on both activities. The experimental group showed significant improvements washing oneself time ($p = 0.038$) (Table 2). There were no between-group differences in the improvement of MBI scores. After 2 weeks of training, participants in the experimental group, rated their self-efficacy in washing oneself as significantly higher [88.8 (SD 14.1) vs. 77.2 (SD 12.8), $p = 0.013$] (Table 3), and they were more satisfied with the training program than participants in the conventional group [97.9 (SD 5.3) vs. 88.3 (SD 13.8)].

Discussion

The aim of this study was to explore the outcomes of applying the seamless self-care training technique. The keystone of this technique is the strengthened collaboration between OTs and nurses in coaching stroke survivors to complete dressing and washing oneself training. The collaboration facilitates the systematic transfer of skills related to the training from a therapy room to a real setting of a patient's daily routine.

The results from this study indicated that this technique could enhance the effectiveness of training and achieving functional gains, a decrease in washing oneself time, and an increase in washing oneself self-efficacy. Although both groups received the same total training time of 14 days, the

Table 1. Demographic data of all 35 study participants

Variables	Experimental group (n=17)	Conventional group (n=18)
Age, years[1]	57.1 (10.3)	59.4 (10.9)
Stroke type[2]		
- Ischemic	15 (88.2)	14 (77.8)
- Hemorrhage	2 (11.8)	4 (22.2)
Side of weakness[2]		
- Right	7 (41.2)	10 (55.6)
- Left	10 (58.8)	8 (44.4)
Dominant hand[2]		
- Right	16 (94.1)	17 (94.4)
- Left	1 (5.9)	1 (5.6)
Gender[2]		
- Male	10 (58.8)	11 (61.1)
- Female	7 (41.2)	7 (38.9)
Duration of stroke, days[3]	16 (12, 35)	26 (16, 58)
Educational level[2]		
- Less than 12 years	10 (58.8)	10 (55.6)
- At least 12 years	7 (41.2)	8 (44.4)
Comorbidities[2]		
- DM	7 (41.2)	9 (50.0)
- HT	15 (88.2)	15 (83.3)
- DLP	12 (70.6)	13 (72.2)
- Heart disease	6 (35.3)	2 (11.1)

[1]Mean (SD), [2]number (%), [3]median (IQR 25, 75)

Table 2. Outcomes of the self-care (dressing and washing oneself) training

Outcomes		Experimental group (n=17)	Conventional group (n=18)	*p*-value[b]
Total self-care score[1]	D1	29.6 (6.7)	31.2 (7.6)	0.519
	D14	39.3 (1.4)	37.2 (3.8)	0.003*
p-value[a]		0.000	0.000	
Normalized gain D14[1]		0.96 (0.1)	0.71 (0.3)	0.005*
High gain[2] (0.7–1)		17 (100)	13 (11.1)	0.028*
Medium gain (0.3 ≤ g <0.7)		0	2 (16.7)	
Low gain (< 0.3)		0	3 (72.2)	
MBI[1]	D1	8.8 (1.6)	7.6 (1.9)	0.036*
	D14	11.4 (0.9)	11.1 (1.3)	0.524
p-value[a]		0.000	0.000	
Washing oneself time, min[1]	D1	8.34 (2.2)	8.12 (3.3)	0.729
	D14	6.44 (1.4)	7.24 (3.2)	0.457
p-value[a]		0.000	0.010	
ΔWashing oneself time D7–D14, min[1]		1.50 (1.4)	0.47 (1.1)	0.038*
Dressing time, min[1]	D1	4.45 (2.6)	2.45 (1.5)	0.022
	D14	3.52 (3.1)	2.21 (1.4)	0.082
p-value[a]		0.001	0.007	
ΔDressing time D7–D14, min[1]		0.52 (0.6)	0.23 (0.3)	0.073

[1]Mean (SD), [2]number (%); [a]*p*-value within group, [b]*p*-value between groups, *significant at $p < 0.05$

Table 3. Dressing and washing oneself self-efficacy

Activity	Day of training	Experimental group (n=17)	Conventional group (n=18)	*p*-value[b]
Washing oneself[1]	D1	72.9 (14.9)	80.0 (14.6)	0.166
	D14	88.8 (14.1)	77.2 (12.8)	0.013*
	p-value[a]	0.0054	0.516	
Dressing[1]	D1	74.1 (15.4)	77.8 (17.0)	0.510
	D14	90.9 (10.3)	82.2 (15.2)	0.065
	p-value[a]	0.0044	0.392	

[1]Mean (SD), [a]*p*-value within group, [b]*p*-value between groups,*significant at $p < 0.05$

experimental group received 3.5 times (14 days vs. 4 days) more practicing time under professional training procedure by OTs and nurses than the conventional group. Because the experimental group received seamless training technique utilizing the self-care checklist for dressing and washing oneself and promoting better communication between professionals, while the conventional group received training in routine approach, not using checklist and no enhanced coordination. As washing oneself is a complicated task requiring more practice than dressing, the seamless self-care training technique was more beneficial for washing oneself. These findings are congruent with the dose-response relationship concept. Participants who performed the tasks more often could obtain more skills and gain more self-efficacy.[7,16] Self-efficacy is an individual's perceived confidence and belief in their capabilities to perform specific tasks or actions and is an important key for successful rehabilitation. Further, self-efficacy is associated with various outcomes, such as physical functioning, including ADL performance, depression, quality of life, and patient satisfaction.[17,18] In addition, Kobyla´nska found that patients who lacked self-efficacy tended to have low functional abilities in post-stroke rehabilitation.[19] Therefore, well-designed training techniques could empower the patients and enhance their sense of competence.

In our study, we created the seamless self-care training technique under an interdisciplinary team working model. Regarding the total treatment time in rehabilitation units, rehabilitation nurses spend much more time with patients compared to OTs.[16] Therefore, we encouraged nursing staff to play an important role as a facilitator for patients when assisting self-care activities instead of their traditional role as a care provider, providing nursing care. A previous study reported that the main reason nurses do everything for patients in almost all activities rather than facilitating them is that the latter is too time consuming. Because of time constraints, especially in the morning, nurses tend to be more concerned about preparing patients to be ready for specific training with other professionals and some nurses perceive that other professionals are predominantly responsible for the rehabilitation processes. Moreover, many patients and families still have expectations that nurses should provide a basic nursing care for them.[9,10,20]

In this study, we measured the time that patients took when trying to do the tasks by themselves. Both groups spent around 10 minutes to complete dressing and washing oneself. From our observation, this only took 5 minutes when performed by nurses in routine care; meaning it took twice as long to achieve the same tasks. Although this study showed the benefit of the seamless self-care training technique, the possibility of putting this approach into practice would need to consider nursing staff's workload. However, another study found that a formal stroke rehabilitation education program could change the interaction between nurses and patients from doing things for them to facilitating them to do things for themselves without any increase in time.[21] Leaders should be strong and coach teams to adhere to goal-oriented outcomes and develop a suitable strategy for their effective time management.[22] On the other hand, if nurses and OTs have a lot of contact time with patients they should have the time to provide patients with meticulous training, which could raise patient satisfaction, which in this study was 9.6% higher in the experimental group than in the conventional group. This might be one factor that could contribute to patients gaining more advanced functional skills. This finding complements the existing research mentioning that higher patient satisfaction is associated with better compliance and outcomes.[22]

The key strength of the seamless self-care training is the greater apparent collaboration between team members than in traditional training, in which no specific time is identified for OTs to work alongside nurses in providing self-care training. With collaboration, the nurse could provide crucial information feedback to the OT, and vice versa, and this could enable them to adapt their care plan accordingly. Moreover, the self-care checklist for dressing and washing oneself was developed in this study to measure the progression of washing oneself and dressing training. This was validated and could be used to aid communication among rehabilitation professionals.

Even though this study showed an improvement in the self-efficacy of participants, a long-term outcome was not followed and a burden on caregivers was not explored. We suggest extending the study into the potential of the seamless self-care training technique to assure its full effects.

In conclusion, this seamless self-care (dressing and washing oneself) training technique is a unique interdisciplinary working model has been proved to be effective. However, translating it into rehabilitation practice could be challenging for changing each discipline mindset for more collaboration and integration.

Disclosure

All authors declare no personal or professional conflicts of interest.

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Appendix

Self-care checklist for dressing and washing oneself
Instruction: Scoring 0 = complete assistance needed, 1 = require verbal cue for completing task, 2= not require assistance
Patient's abilities in washing oneself and dressing
Taking off the button shirt
1. Slightly take shirt off affected shoulder
2. Take shirt off unaffected shoulder
3. Pull shirt off unaffected arm
4. Pull shirt off affected arm
Total score
Taking off pants (choose either A or B which compatible with patient's ability)
A. In standing position (for a patient who needs minimal assistance in tranferring)
1. Stand up and pull pants down to knee level
2. Sit at the edge of the bed
3. Remove pants from unaffected leg
4. Remove pants from affected leg
B. In supine position (for a patient who needs moderate assistance in tranferring)
1. Lie on one side or raise the hip in supine position then pull pants down to knee level
2. Sit at the edge of the bed
3. Remove pants from unaffected leg
4. Remove pants from affected leg
Total score
Putting on pants (choose either A or B which compatible with patient's ability)
A. In standing position (for a patient who needs minimal assistance in tranferring)
1. Sit and cross affected leg over unaffected leg
2. Put pants on affected leg then pull pants up above knee level
3. Put pants on unaffected leg
4. stand up and use unaffected arm to pull pants up to waist level
B. **In supine position (for a patient who needs moderate assistance in tranferring)**
1. Sit and cross affected leg over unaffected leg, put pants on affected leg
2. Put pants on unaffected leg then pull pants up as high as possible
3. Lie down on bed in supine position
4. Lie on one side or raise the hip in supine position then use unaffected arm to pull pants up
Total score
Putting on shirt
1. Properly lay down the shirt to put on
2. Put the shirt on affected arm
3. Use unaffected hand to pull the shirt up to the shoulder, move the rest of the shirt behind the body
4. Put the shirt on unaffected arm
Total score
Washing oneself
1. Turn on the faucet, use shower head/water dipper to wet the entire body
2. Rub the soap onto entire body, with or without assistive devices
3. Wash the soap from the body thoroughly
4. Dry the body thoroughly
Total score
Sum score

Original Article

Effects of Applications on Computer Tablet for Cognitive Training in Stroke Patients

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ABSTRACT

Objectives: To study the effects of a computer tablet applications for cognitive training on cognitive functions in stroke patients.
Study design: A quasi-experimental, one group, pretest-posttest design.
Setting: Occupational Therapy Unit of Thammasat University Hospital
Subjects: Fifteen patients with first stroke and cognitive impairment, aged between 35-70 years, and duration of stroke less than a year, who received occupational therapy service from December 2018 to June 2019
Methods: Participants were selected by purposive sampling. The mini-mental state examination (MMSE)-Thai 2002 was used to screen for cognitive impairments. Participants received a tablet computer-based cognitive training under supervision of an occupational therapist 3 times per week for 6 consecutive weeks. They underwent the Digit Span Test, the Thai Cognitive-Perceptual Test (Thai-CPT) and the Dynamic Loewenstein Occupational Therapy Cognitive Assessment (DLOTCA) before and after the training period. Descriptive statistics and non-parametric Wilcoxon Signed Ranks Test were used to analyze data.
Results: The average age (SD) of the participants was 58 (11.39) year old. The male:female ratio was 1.1:1. The improvement of attention, memory and executive functions reached statistical significance ($p < 0.05$).
Conclusion: The tablet applications for cognitive training which were used in this study improved attention, memory and executive functions in stroke patients.

Keywords: cognition, perception, computer tablet, stroke rehabilitation, occupational therapy

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Introduction

Stroke is a significant health problem world-wide. The World Health Organization (WHO) reported global death rate from stroke as 17.9 million per year, which is 77% in 100,000 people especially in those who were 70 years old and over. It is the second cause of death to coronary disease.[1] In Thailand the current stroke situation reported by the public health statistics shows 47.15% death rate in 100,000 people and is the number one cause of death in non-communicable diseases (NCDs), and there are 30,837 patients diagnosed per year, and the number may increase if correct prevention is not established.[2] One in three stroke patients are permanently disabled and 20% needed long-term care in rehabilitation clinics. The residual disability affects their quality of life, self-image and their families' economy.[3] Stroke patients have impairments in the motor and sensory systems which cause difficulties in body movements. In addition, many stroke survivors have cognitive impairments from lesions in different areas of the brain, which become barriers to being independent. They must depend on their families and the society which leads to social disadvantages.[4] Cognitive impairments after stroke is prevalent and affects 72% of all stroke patients in the first 1-3 months.[5] The most commonly impaired domains are the followings: attention, memory, language, orientation, and executive functions.[6] Generally, maximal recovery is within 12-18 months and 30% of the patients who still have cognitive impairments after one year develop post-stroke dementia.[7] Cognitive rehabilitation models commonly used in stroke patients in Occupational Therapy clinics are 1) remedial or restorative approach which stimulates neuroplasticity using table top or computer-based activities with other specific techniques by professionals, and 2) adaptive or compensatory approach which improves patients ability to engage in occupations under their limitations and compensate for loss abilities through activities analysis and synthesis, cognitive strategies, and environment modifications and adaptations.[8]

Recently, several computer-based cognitive trainings have become available for use in clinical practice such as computerized software, tablets, gaming consoles and virtual reality systems. Prior oversea studies on the effects of computer-based cognitive training varied in program formats, measurement of results, and research methodologies.[9] Several studies in stroke patients showed that a computer assisted cognitive training can improve working memory, delayed

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memory, attention span, visual and auditory learning, verbal fluency, and depressive symptoms.[10,11] It was suggested that supervision by a qualified therapist who could modify level of task difficulty to suit individual patient at their respective stage of recovery, is another very important factor.[12,13]

Even though studies about computer-based cognitive trainings from oversea are numerous. However, the number of studies from Thailand is still limited. Cultural bias is a potential factor which limits the validity of adopting training software which were developed abroad. Recently in Thailand, a CogTA-Tab for cognitive training has been developed and designed based on Thai contexts.[14] Researchers chose to use a tablet as therapy media because modern tablets are so portable, easy to operate, have fast and powerful computing power and yet do not cost a fortune. Stroke patients, who typically experience weakness on one side of the body could without much difficulty could carry the tablet around and operate them with little assistance. The goal of this research was to study the effect of cognitive training using this software on tablet computer by measuring changes of attention, memory, and executive functions after a course of tablet-based training with the CogTA-Tab in a group of stroke patients.

Methods

This research was approved by the Research Ethics Committee of the Faculty of Associated Medical Sciences, Chiang Mai University No. AMSEC–61EX-021 and the Human Ethic Sub-committee of Thammasat University, No. 3 (Faculty of Health Science and Science and Technology), the project No. 197/2561. This research was supported and funded by the Faculty of Associated Medical Sciences, Chiang Mai University, through 2018 annual research funds. This research was a quasi-experimental, one group, pretest-posttest design.

Participants

Stroke patients who received occupational therapy at Thammasat University Hospital from December 2018 to June 2019.

Inclusion Criteria

- First-time stroke patients, during post-acute phase with less than one year after onset.
- Age between 35-70 years old.
- Having cognitive impairments screened with the Mini-Mental State Examination Thai Version–2002 (MMSE–Thai 2002). Score cut point was ≤ 14 for those who did not enter schools or were unable to read, at 17 for those who passed primary school, and ≤ 22 for those who had higher education.[15]
- Able to read and write in Thai
- Being familiar with using a computer, a tablet or a smartphone.

Exclusion Criteria

- Having visual impairment or communication limitation, dementia or other mental disorders.

Withdrawal criteria

- Receiving less than 80% of the training sessions.
- Taking medications affecting cognitive abilities during the training.

Sample size calculation

Based on data from a previous study,[16] sample size calculation was made with a computer software G*power 3.1. Sample size of 13 were needed when the following parameters were defined: effect size 1.01 with .05 deviation and .08 Power of Test. To prevent drop outs, the researchers added 2 more participants, therefore this research sample size was a total of 15 participants.

Outcome measurement

1. The Forward and Backward Digit Span Test were used to assess attention in this research.[17]
2. Memory was assessed with the “Recall” and “Recognition” subtest of the Thai Cognitive-Perceptual Test (the Thai-CPT).[18]
3. For executive functions, the Matching and Categorization, and the Problem-Solving subtest of the Thai-CPT were used to assess executive functions. An assessor received a formal training in order to use and score this tool accurately.[18] In addition, four topics from two subtests: 1) Visuomotor Construction: Plain Block Design and Puzzle, and 2) Thinking Operation: Pictorial Sequence A & B to assess executive functions, from the Dynamic Lowenstein Occupational Therapy Cognitive Assessment for Adult (the DLOTCA) were also used.[19]

Intervention

Application on Tablet for Cognitive Skills Training in Stroke Patients

“The CogTA-Tab” is a computer program developed by Munkhetvit et al. under cognitive rehabilitation models commonly used in occupational therapy practice.[14] It focuses on training 3 domains of cognitive skills affected by stroke: attention, memory and executive functions. Its application was developed with the software development tool “Unity3D” Version 2018. It can be installed on smartphones or PC tablet which operates on Android operating system Version 5.0 and up. The hardware used in this study was a Huawei Media Pad T5 with 10 inches screen. The CogTA-Tab consists of 15 cognitive training games as follows: 1) Attention training games, in which a visual target randomly appears various locations on screen, then patients respond by touching the screen where the targets are until they have collected the set number of targets. These games require visual attention and orientation in order to search for the targets. Example of the activities are maze game, savings coins, finding different numbers, finding animals, fishing game, provincial explorer, and calm touching game (shown in Figure 1 and 2) Memory training games that feed sensory information including

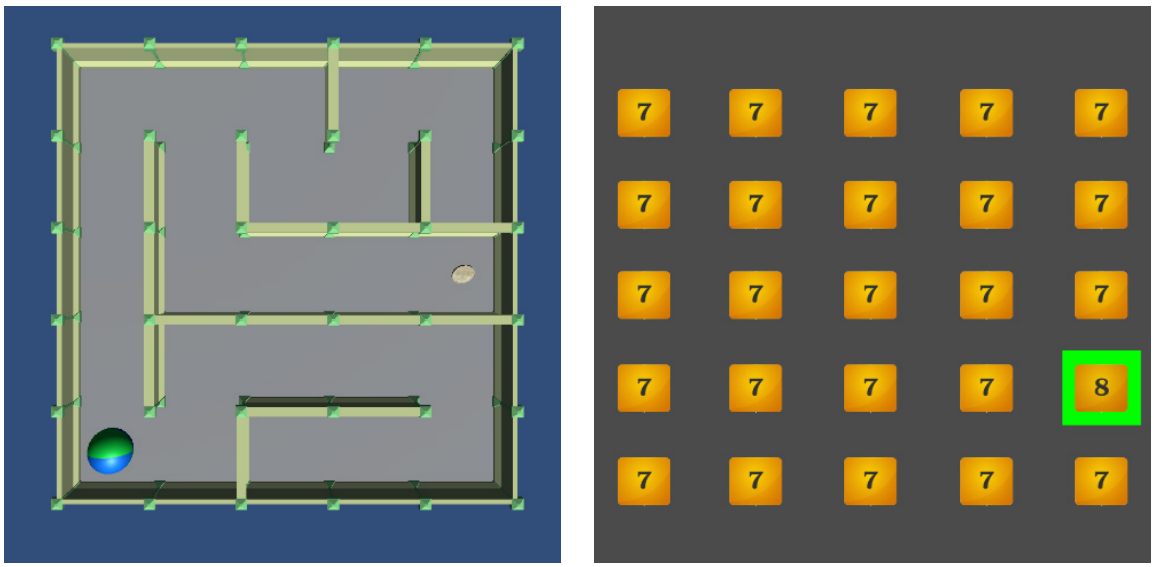

Figure 1. A) Sustained attention training in games maze, B) Focused attention training in find the different number games

Figure 2. Recall memory training on tablet

sounds, pictures, numbers and symbols to stimulate patients to process the information from hearing and seeing senses, then respond by collecting short-term memory and recall memory. Example of the activities are number echo, count lyrics words, can you remember? and listen carefully game. (shown in Figure 2 and 3) Executive functions training games which stimulate patients to use executive functions through pictures, words, and numbers. The patients must try to organize, sort, plan ahead, show flexible thinking and abstract thinking, and stimulate patients to complete the targeted activity. The activities are the train, don't be confused, picture mystery game, and matching pictures game. Within each type of games, there are 3-4 levels of difficulties. Each patient will start at the easiest level (Level 1), then the level of difficulty will gradually increase in each training. In one session, the patients receive training of all these 3 cognitive skills. Moreover, before the training program, the participants and their caregivers received education about stroke and cognitive impairments following stroke. During each 45-minute sessions individualized training were delivered under direct supervision by the researcher. During the 6 consecutive weeks, each patient received 18 treatments in addition to their conventional occupational therapy rehabilitation program at Thammasat University Hospital.

Steps of the study

1. After receiving human research approval from the three Research Ethics Committees mentioned-above, the researchers started collecting 15 participants via specific sampling, explained the research objectives, intervention and protection rights, and asked them to sign an informed consent form.
2. The trained assessor used the Digit Span Test, the Thai CPT, and the DLOTCA to assessed the participants' cognitive functions before the training program (pre-test).
3. Each participant received cognitive skills training using tablet application for 45 minutes per session, 3 sessions per week for 6 consecutive weeks, in combination with a conventional occupational therapy program at Thammasat University Hospital.
4. After completing the cognitive training program, the same assessor reassessed all participants by using the same tools.
5. Data were analyzed and interpreted.

Statistical analysis

Statistical analysis was done by computer program SPSS version 22.0. Demographic data were described as number, percentage, mean and standard deviation (SD). The cognitive functions scores before and after the intervention were analyzed using non-parametric statistics, Wilcoxon Signed Ranks Test where statistical significance was determined at $p < 0.05$.

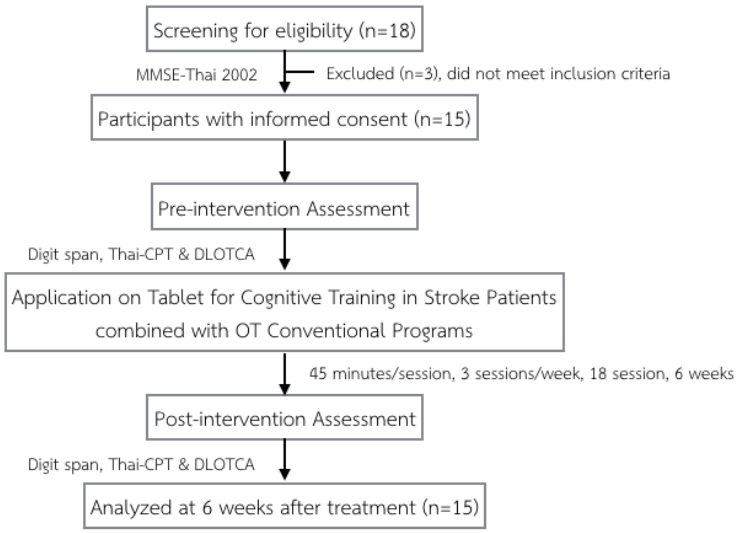

Figure 3. Flow of the study

Results

Fifteen participants were recruited: eight were males (53.3%), average age was 58.73 (SD 11.39; range 35-70) years, 86.7% received education higher than primary level; average duration of stroke was 4.53 months, 80% had ischemic stroke, and 53.3% experienced right-side weakness; and 66.7% were right-handed. Using the MMSE-Thai 2002, the participants' cognitive screening results had an average score of 19.80 (as shown in Table 1).

As demonstrated in Table 2, majority of the participants had improved cognitive test scores (positive rank) in all three cognitive skills. There was no participant with lower test scores after training (negative rank), and there were only small number of participants with same scores despite of training (Ties). Statistical analysis comparing before and after training cognitive measurements with Wilcoxon Signed Ranks Test confirmed significant improvement in attention, memory and executive functions at $p < 0.05$, except for recognition memory. The mean rank of each test score both before and after training, including number of participants with negative ranks, positive ranks and tie, as well as the corresponding statistic significant level can be seen in Table 2

We also observed that stroke patients who had weakness in their dominant arm and hand had difficulties getting used to functioning with their non-dominant hand to respond on the tablet screen. This was common in the initial phase of training using tablet application. Another common problem for older age stroke patients was difficulty using their fingertips to touch the screen due to skin dryness. The use of touch screen stylus could overcome this problem.

Discussion

This research found that the cognitive training program using the CogTA-Tab application on a tablet improved cognitive functions in stroke patients significantly ($p < 0.05$) which proved the research hypothesis that this tablet application would help improve attention, memory and executive functions in all participants after receiving the training. This research shows positive effects of cognitive training programs conforming to the research by Zucchella et al. which combined multi-domain computer-based cognitive rehabilitation program and metacognition strategies training as an early intervention for patients with stroke during subacute phase.[20] The study done by Zucchella et al. used a retraining approach in the belief that repetitive method of skills training or computer-based exercises help to improve cognitive functions in stroke patients, and showed that patients have improved skills in visual attention and verbal memory after 4 weeks of training and suggested that logical-executive functions and the patients' quality of life may take longer

Table 1. Demographic and clinical characteristics of the study participants (n=15)

Demographic data	
Gender[1]	
Male	8 (53.3%)
Female	7 (46.7%)
Age (years)[2], min=35 years, maximum=70 years	58.73 (11.39)
Education level[1]	
Primary school and lower	2 (13.3%)
Higher than secondary school	13 (86.7%)
Duration of stroke (month)[2]	4.53 (2.94)
Types of stroke[1]	
Ischemic	12 (80%)
Hemorrhagic	3 (20%)
Affected side[1]	
Right hemiparesis	8 (53.3%)
Left hemiparesis	7 (46.7%)
MMSE – Thai 2002 score[2]	19.80 (1.69)

[1]Number (%), [2]mean (SD)
*MMSE-Thai 2002, Mini-Mental State Examination-Thai Version 2002

Table 2. Mean rank, z-score, p value between pre-test and post-test scores

Cognitive skills	Negative ranks (post < pre)		Positive rank (post > pre)		Ties (post = pre)	z	*p*-value
	n=15	Mean rank	n=15	Mean rank			
Attention							
Forward digit span	0	0.00	12	6.50	3	-3.089[a]	0.002*
Backward digit span	0	0.00	14	7.50	1	-3.359[a]	0.001*
Total	0	0.00	14	7.50	1	-3.329[a]	0.001*
Memory							
Recall	0	0.00	13	7.00	2	-3.192[a]	0.001*
Recognition	0	0.00	3	2.00	12	-1.604[a]	0.109
Total	0	0.00	14	7.50	1	-3.314[a]	0.001*
Executive functions							
Matching & categorization	0	0.00	8	4.50	7	-2.640[a]	0.008*
Problem solving	0	0.00	14	7.50	1	-3.346[a]	0.001*
Visuomotor construction &thinking operation	0	0.00	14	7.50	1	-3.318[a]	0.001*
Total		0.00	15	8.00	0	-3.415[a]	0.001*

*Statistically significant difference in means ($p < 0.05$), a based on negative ranks

to restore to show more apparent change.[20] Similar to the study by Westerberg et al., memory training using a personal computer at home in combination with daily online follow-up by therapist could improve working memory and attention in stroke patients, and the program should provide graded activities from easy to hard levels to suit each patient's ability, as well as to provide follow-up or feedback from therapist to maintain a continuity of training and patients' adherence to training at home.[21] Moreover, the study by Prokopenko et al. used computer correction in cognitive training by repetitive of correction to help stimulate perception, restoration, and processing of information in the brain. It was found that stroke patients had increased skills in attention, visual and spatial memory after the training.[22]

The CogTA-Tab application used in this study consisted of large variety training activities which may promote active engagement of participants during training. Uses of pictures, sounds, colors and contents related to Thai context might also be another factor that supports the effectiveness of this particular software. In addition, a direct personal supervision during the training allowed an immediate positive feedback which further motivated the participants and retained consistently high patient effort level during training. Furthermore, it allowed for negative feedback, which increased the challenge and motivated the participants to develop their capacity. The positive relationship between the trainer and the participants leaded to relaxed atmosphere during the training which could facilitate higher cognitive performance. It is well known that cognitive strategies training using internal and external strategies, errorless learning technique and cueing or prompting techniques by therapist, could promote development of executive functions, as well as generalization of the new to other activities.[23,24] During this research, the use of the application on tablet for specific cognitive skills training were carried out according to the principles of cognitive restorative approach and applied graded activity, tailored to individual pretraining ability level.

We believe that such improvements might come from the fact that the brain structures and functionals change after received the training program. Lin et al. studied the recovery mechanism of brain functions in stroke patients who received computer-based cognitive training programs to restore executive functions and memory, and found significant structural changes in the brain function networks especially in the hippocampal, the frontal and the left parietal lobe areas, which relate to memory and executive functions.[25] In a study by Yang et al. found that after integrated computerized cognitive training program, the stroke patients showed increased of the hippocampal functional connectivity mainly located in the prefrontal gyrus and medial temporal areas which are related to improved memory and change in brain structures during default mode network (DMN) controlled by the parietal lobe of the brain in posterior cingulate, ventral medial prefrontal, and dorsal medial prefrontal cortex, which are associated of cognitive functions.[26] Meanwhile, Yeh et al. studied cognitive skills training with aerobic activity using computer program and believed that it might speed up the process of recovery in neural and brain cells in affected and nearby areas, help increase axons for synaptic connectivity which affected the brain functionals as seen in fMRI, and the changes are more obvious during 3-6 months after regular training.[27] According to the study of Zhang et al., the effects of a computerized multi-domain cognitive training run on a tablet device on improving skills in reasoning, memory, visuospatial skill, language, calculation, and attention were changes in the brain's gray matter volume in the right angular gyrus and parietal area, near the intra-parietal sulcus, in patients with amnestic mild cognitive impairment.[28]

Our study showed significant changes in the participants' cognitive skills in attention, executive functions and recall memory after completing the training program. However, there was no significant difference in recognition memory between pre- and post-test intervention. We assumed that this is the phenomenon called ceiling effect resulting from the very high to full pre-test score in recognition memory. It is in accordance with the study by Chaiwong et al. finding no difference in recognition memory between pre- and post-test in elderly with mild cognitive impairment.[29] This may also be due to the level of difficulty of the assessment tool, which gives the high pre-test scores. Thus, after the intervention, those whose the pre-test scores were high, shows no difference. Bastin et al. explained a hypothesis that patients whose hippocampal area of the brain was damaged will lose the ability to recall or recollection memory.[30] However, the patients will still have the ability of recognition depending on the autonomic familiarity and personal experiences. Thus, this research found no difference in recognition as the most patients remaining abilities.

The limitations of this research were a small sample size and only one sample group. Some activities in the CogTA-Tab application are similar to some test items from the assessment tools, and could result in more or less practice effects. Further study can be designed to overcome these by including a control group with adequate number of subjects, as well as choosing outcome measurements that did not overlap with the training methods.

In conclusion, using the CogTA-Tab application developed by Thai researchers for 6 weeks of cognitive training could improve cognitive functions in stroke patients during post-acute phase.

Disclosure

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Original Article

Prevalence and Factors Related to Post Stroke Shoulder Pain in Hemiplegic Patients Receiving Home-based Rehabilitation in Post-acute Phase

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ABSTRACT

Objectives: To study the prevalence of shoulder pain and related factors in stroke patients with home-based rehabilitation in the post-acute phase.
Study design: Cohort study.
Setting: Phatthalung Hospital, Thailand.
Subjects: Stroke patients who had a home-based rehabilitation program and monthly follow-up for 6 months at Rehabilitation outpatient clinic from February 2019 to January 2020.
Methods: Demographic and medical data including types of stroke, motor power, sensation, the Brunnstorm's arm recovery stage, shoulder subluxation, shoulder pain, self-care activities and home exercises, were recorded. Causes of shoulder pain was identified and treated until resolved. Factors such as using an overhead pulley for range of motion exercise and not using a shoulder sling while walking, were evaluated and analyzed.
Results: Of 110 patients, 58% reported shoulder pain, mean age was 64.14 (SD 13.91) years old, 80.9% had ischemic stroke, 58% had left side weakness, 52.7% had impaired sensation, 41.8% had the Brunnstorm's arm recovery stage 1, and 50% had shoulder subluxation. Of those with shoulder pain, 64.1% developed shoulder pain within a month after onset. The most common diagnosis of shoulder pain was impingement syndrome (57.8%). Impaired sensation (adjusted OR=17.52; 95% CI 4.74-64.78), using an overhead pulley for range of motion exercise (adjusted OR = 33.92; 95% CI 3.35-343.39), and walking without a shoulder sling (adjusted OR = 11.68; 95% CI 1.28-106.88), were significantly associated with post stroke shoulder pain ($p < 0.05$).
Conclusion: Post stroke shoulder pain was common among patients with home-based rehabilitation during post-acute phase. Factors significantly associated with shoulder pain were having an impaired sensation, using an overhead pulley for shoulder range of motion exercise, and not using a shoulder sling while walking. Patient education including using a shoulder sling and not using an overhead pulley for shoulder exercise at home exercise should be emphasized to prevent shoulder pain.

Keywords: shoulder pain, shoulder subluxation, stroke, exercise, home-based rehabilitation

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Introduction

Stroke, a common disease worldwide, usually causes brain damage or hypoxia resulting in an abnormal nervous system such as impaired motor control and tone change. Shoulder muscles weakness and decreased muscle tone in acute phase lead to shoulder subluxation and pain in which consequently leads to joint stiffness, limits movement and function of the hemiplegic arm, decreases activities of daily living (ADL) ability and increases the duration of treatment and rehabilitation.[1,2]

The incidence of shoulder pain has been reported to be 5%-84% depending on the duration of stroke and study populations.[3-6] In a previous study on stroke during post-acute inpatient rehabilitation in Thailand, 19% of the patients reported shoulder pain and more in those with shoulder subluxation.[3] A cohort study in the UK found that 40% of patients developed post stroke shoulder pain on the affected side and it was strongly associated with abnormal shoulder joint examination, ipsilateral sensory abnormalities and arm weakness.[6]

In Thailand, early but short hospital-based rehabilitation is provided to patients with stroke disabilities to reduce hospital cost.[3] After discharge, they have to continue with either a home-based rehabilitation or a community-based rehabilitation program. Most of disabled patients and their families tend to opt for home-based rehabilitation because of environmental familiarity, convenience and ease of continuity of care. However, there was no data on incidence of post stroke shoulder pain in stroke patients receiving home-based rehabilitation in Thailand.

Based on our observation, a large number of patients came back with shoulder pain in the first month after discharge. We observed that instead of passive ROM done by caregiver, some sort of equipment of range of motion exercise such as an overhead pulley was used. In addition, it has been observed that there is an overhead pulley installed at nearly all community rehabilitation centers and many health volunteers still advise patients to use it for shoulder exercise.

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Therefore, the purpose of this study was to determine the prevalence of shoulder pain and associated factors in the hemiplegic patients practicing home-based rehabilitation, and whether an overhead pulley for shoulder exercise related to shoulder pain. The results of this study would help improve a home-based as well as a community-based rehabilitation program to prevent post stroke shoulder pain that interferes with ADL and thus reduces quality of life.

Methods

Participants

Stroke patients who were discharged from the stroke unit and had been appointed to follow-up within 1 month of stroke onset at Rehabilitation Outpatient Clinic, Phatthalung Hospital, Thailand.

Inclusion criteria

- Older than 18 years of age
- Less than a month from stroke onset
- Motor power of the hemiplegic arm less than grade 5

Exclusion criteria

- Could not communicate.
- Lost to follow up

Study protocol

1. After receiving the hospital Ethical Review Board's approval, those who met the inclusion criteria were asked to participate in the study.

2. After the patients signed the informed consent, they were registered and the following baseline variables: age, gender, main type of stroke, comorbid disease, and stroke onset date were collected. Physical and neurological examinations were performed to assess shoulder pain, shoulder subluxation, motor power, sensation and the Brunnstorm's arm recovery stage.

3. The patients were interviewed by predetermined structured questionnaires consisting of history of shoulder pain and injury before stroke onset. In addition, they were asked how often they did range of motion (ROM) exercise, whether or not they used an overhead pulley and wore a shoulder sling while walking.

4. If shoulder pain presented, treatments for shoulder pain such as physical modalities and medications were prescribed. Physical modalities included heat therapy and transcutaneous electrical nerve stimulation (TENS). Medications such as analgesics, anti-inflammatory drugs, antispastics, anti-neuropathic pain medications, were prescribed. Oral corticosteroid was only for complex regional pain syndrome. Shoulder pain was re-assessed every month until resolved or 6 months after onset. The following information: diagnoses of pain, treatments, treatment outcomes, numerical rating scale (NRS) and duration of pain, were recorded.[7,8] If the patients did not have shoulder pain, they were followed up monthly for 6 months.

Statistical analysis

Descriptive statistics was used to demonstrate the characteristics of the subject demographics. All variables were examined with a univariate analysis and all variables with a $p < 0.05$ were included in a multivariate logistic regression analysis. Multivariate analysis was performed by binary logistic regression using forward analysis (variables were included and excluded from the model using a cut-off p of 0.05). All analyses were performed with SPSS version 17 for Windows.

Results

There were 142 patients who underwent an initial interview. During the 6-month follow up period, 31 failed to follow up and 1 died, resulting in the remaining 110 patients recruited in the study. The demographic baseline of 110 patients is shown in Table 2: 59 patients (53%) were males, age ranged from 33 to 98 years with the mean of 64.14 (SD 13.91) years, 80.9% had ischemic stroke, 74.5% had underlying hypertension, 29.1% had diabetes, 68.2% had dyslipidemia, 8.2% had heart disease and 4.5% had previous stroke; 58% had left side weakness, 52.7% had impaired pinprick sensation, 41.8% had the Brunnstorm's arm recovery stage 1, and 40% had motor power grade 0; 50% had shoulder subluxation and 58% experienced post stroke shoulder pain.

Factors related to shoulder pain are shown in Table 3. There was no statistically significant relation to age, type of stroke, gender, comorbid disease, or weak side. Upon a simple regression analysis, statistically significant association was observed between post stroke shoulder pain and shoulder subluxation (crude OR = 4.08;95%CI 1.01-1.85), motor power

Table 1. Terms, definition and available references

Terms	Definitions	References
Post stroke shoulder pain	Shoulder pain on the same side of hemiparesis	
Shoulder subluxation	Having a palpable gap between the acromion and humeral head caused by a change in the mechanics of the glenohumeral joint.	Suethanapornkul et al[3]
Impaired sensation	When using a pinprick on the face, arms, trunk and legs and comparing the sensation sharpness side by side, the difference is mild to moderate or severe to total loss is observed.	Lyden[9]
Walking without a shoulder sling	Not wear a sling during walking.	Jeong et al[10]
Not exercising everyday as instructed	Not exercise to maintain range of motions of the shoulder regularly every day.	Winstein et al[11]

Table 2. Demographic and neurological examination data of all 110 participants at baseline

Demographic variables	Number (%)
Sex: male	59 (53.6)
Age (years)	
≤ 60 years	47 (42.7)
> 60 years	63 (57.3)
Type of stroke	
Infarction	89 (80.9)
Hemorrhage	21 (19.1)
Comorbid diseases	
Hypertension	82 (74.5)
Diabetes	32 (29.1)
Dyslipidemia	75 (68.2)
Heart disease	9 (8.2)
Previous stroke	5 (4.5)
Side of weakness	
Right	46 (41.8)
Left	64 (58.2)
Brunnstorm's arm recovery stages	
1 Flaccidity	46 (41.8)
2 Synergies, some spasticity	18 (16.4)
3 Marked spasticity	16 (14.5)
4 Out of synergies, less spasticity	9 (8.2)
5 Selective control movement	15 (13.6)
6 Isolated movement	6 (5.5)
Motor power (elbow flexors of the affected side)	
0 Total paralysis	44 (40.0)
1 Palpable or visible contraction	8 (7.3)
2 Active movement, gravity eliminated	27 (24.5)
3 Active movement, against sum resistance	11 (10.0)
4 Active movement, against full resistance	20 (18.2)
Sensation	
Normal	52 (47.3)
Impaired	58 (52.7)
Shoulder subluxation, yes	55 (50.0)

grade 0 (crude OR = 3.25; 95%CI 1.07-9.82) and grade 2 (crude OR = 4.41; 95%CI 1.28-15.17), the Brunstorm's arm recovery stage 1-2 (crude OR = 3.33; 95%CI 1.20-9.26), impaired sensation (crude OR = 13.43; 95%CI 5.30-34.04), using an overhead pulley for shoulder exercise (crude OR = 23.57 ;95%CI 3.04-182.67), and walking without wearing a shoulder sling (crude OR = 9.17; 95%CI 1.52-55.34) ($p < 0.05$).

The multivariate logistic regression analysis identified impaired sensation (adjusted OR = 17.52; 95% CI 4.74-64.78), ROM exercise with an overhead pulley (adjusted OR = 33.92; 95% CI 3.35-343.39) and walking without a shoulder sling (adjusted OR = 11.68; 95% CI 1.28-106.88) to be associated with post stroke shoulder pain (Table 4). The following factors: age, gender, type of stroke, underlying diseases, side of weakness, motor power, shoulder subluxation and the Brunnstorm 's arm recovery stage, were not significantly associated with post stroke shoulder pain.

Of the 64 patients with shoulder pain, 64.1% developed shoulder pain within a month, 17.2% within 2 months and 8 (12.5%) within 3 months after the stroke onset. The stroke duration of patients with shoulder pain was 1.63 (SD 0.98) months on average. The diagnoses of post stroke shoulder pain were obtained from history taking and physical and neurological examinations as shown in Table 5. All patients were treated with treatments such as ranges of motion exercise, modality, non-sterioidal anti-inflammatory drugs (NSAIDs), oral steroid for complex regional pain syndrome (CRPS), neuropathic pain drugs. Most patients had good outcomes: 54.7% had completely resolved of shoulder pain, and 21.9% had some improvement (pain scores 1 to 3) but 23.4% still had shoulder pain (pain scores 4 to10) at 6 months after the onset of the stroke. The duration of shoulder pain was 3.22 (SD 1.45) months on average.

Discussion

This study revealed that the occurrence hemiplegic shoulder pain was 58%. In the literature, prevalence of shoulder pain varied and ranged from 5 to 84%.[3-6] In a study of post-acute stroke rehabilitation in Thailand,19% of admitted patients had hemiplegic shoulder pain[3] which was much less than this study. The difference in the prevalence of shoulder pain might be due to different settings as well as activities and exercises at home. The results of this present study revealed that those walking without a shoulder sling developed shoulder pain significantly. Ambulatory patients with a flaccid shoulder muscles need a support, such as a shoulder sling, to decrease the severity of shoulder subluxation by narrowing the gap between acromion and the head of humerus and realigning scapular symmetry.[10,12] Shoulder slings would protect the hemiplegic arm and hand from trauma and prevent traction of brachial plexus and shoulder capsule, but not recommended when muscle tone increased.[10] One possible reason that the patients did not use a shoulder sling was that they could not put on the sling by themselves and need a caregiver to assist them.

This present study also demonstrated that patients with impaired pinprick sensation had a high risk of shoulder pain. Impaired pain perception makes shoulder of hemiplegic patients more prone to injury.[13] The brain lesion itself or reduction in mobility and sensory input might lead to an imbalance in central neural control of the sympathetic system.[4] In addition, inappropriate handling, positioning and transferring could exert great stress on the vulnerable shoulder.[14] The problem may be exacerbated by sensory and perceptual deficits. However, this study did not evaluate improper handling and transferring techniques used in the patients.

Moreover, there was no relation between shoulder pain and age, sex, type of stroke, underlying disease, side of weakness that agree with other studies.[3-6] During the study, the patients were instructed to do shoulder ROMs exercises one session per day to maintain ROMs and prevent joint problem. However, the daily ROM exercises was not related to hemiplegic shoulder pain. It is suspected that shoulder

Table 3. Factors related to post stroke shoulder pain: univariate analysis (n=110)

Factor		Shoulder pain		Crude OR	*p*-value	95% CI
		Yes	No			
Gender						
Female		29 (56.9)	22 (43.1)			
Male		35 (59.3)	24 (40.7)	1.11	0.794	0.52-2.37
Age						
≤ 60 years		26 (55.3)	21 (44.7)			
> 60 years		38 (60.3)	25 (39.7)	1.23	0.599	0.57-2.64
Type of stroke						
Infarction		48 (53.9)	41 (46.1)			
Hemorrhage		16 (76.2)	5 (23.8)	2.73	0.070	0.92-8.11
Comorbid disease						
Hypertension	No	17 (60.7)	11 (39.3)			
	Yes	47 (57.3)	35 (42.7)	1.15	0.753	0.48-2.76
Diabetes	No	50 (64.1)	28 (35.9)			
	Yes	14 (43.8)	18 (56.3)	0.44	0.052	0.19-1.00
Dyslipidemia	No	23 (65.7)	12 (34.3)			
	Yes	41 (54.7)	34 (45.3)	0.63	0.276	0.27-1.45
Heart disease	No	58 (57.4)	43 (42.6)			
	Yes	6 (66.7)	3 (33.3)	1.48	0.592	0.35-6.26
Previous stroke	No	61 (58.1)	44 (41.9)			
	Yes	3 (60.0)	2 (40.0)	1.08	0.933	0.17-6.75
Side of weakness						
Right		24 (52.2)	22 (47.8)			
Left		40 (62.5)	24 (37.5)	1.59	0.280	0.71-3.30
Brunnstorm's arm recovery stages						
1-2		43 (67.2)	21 (32.8)	3.33	0.021*	1.20-9.26
3-4		13 (52.0)	12 (48.0)	1.76	0.347	0.54-5.73
5-6		8 (38.1)	13 (61.9)			
Motor power						
0		28 (63.6)	16 (36.4)	3.25	0.037*	1.076-9.82
1		5 (62.5)	3 (37.5)	3.10	0.193	0.57-16.96
2		19 (70.4)	8 (29.6)	4.41	0.019*	1.28-15.17
3		5 (45.5)	6 (54.5)	1.55	0.568	0.35-6.94
4		7 (35.0)	13 (65.0)			
Sensation						
Normal		15 (26.8)	37 (71.2)			
Impaired		49 (84.5)	9 (15.5)	13.43	<0.001*	5.30-34.04
Subluxation						
No		23 (41.8)	32 (58.2)			
Yes		41 (74.5)	14 (25.5)	4.08	0.001*	1.81-9.15
Shoulder pain or injury before stroke onset						
No		57 (55.9)	45 (44.1)			
Yes		7 (87.5)	1 (12.5)	5.53	0.116	0.67-46.57
Overhead pulley used						
No		42 (48.3)	45 (51.7)			
Yes		22 (95.7)	1 (4.3)	23.57	0.002*	3.04-182.67
Walking without shoulder sling						
No		9 (64.3)	5 (35.7)			
Yes		33 (94.3)	2 (5.7)	9.17	0.016*	1.52-55.34
Not exercising every day as instruction						
No		37 (41.6)	52 (58.4)			
Yes		9 (42.9)	12 (57.1)	1.05	0.915	0.40-2.76

Number (%), * statistically significant at $p < 0.05$

Table 4. Multivariate logistic regression result of risk factors for development of shoulder pain

Factor	Yes	No	Crude OR	Adjusted OR	*p*-value	95% CI
Sensation						
Normal	15 (26.8)	37 (71.2)				
Impaired	49 (84.5)	9 (15.5)	13.43	17.52	< 0.001*	4.74-64.78
Exercise with an overhead pulley						
No	42 (48.3)	45 (51.7)				
Yes	22 (95.7)	1 (4.3)	23.57	33.92	0.003*	3.35-343.39
Walking without shoulder sling						
No	9 (64.3)	5 (35.7)				
Yes	33 (94.3)	2 (5.7)	9.17	11.68	0.030*	1.28-106.88

Number (%), *statistically significant at $p < 0.05$

Table 5. Diagnoses, treatments and outcome of post stroke shoulder pain (n=64)

	Number (%)
Diagnoses	
Adhesive capsulitis	17 (26.6)
Shoulder impingement	37 (57.8)
Complex regional pain syndrome	15 (23.4)
Central pain	16 (25.0)
Others (bicipital tendinitis, acute arthritis, severe spasticity)	5 (7.8)
Treatments	
ROM exercise	60 (93.8)
Modality	17 (26.6)
Oral steroid	15 (23.4)
Neuropathic pain drug	23 (35.9)
Non-steroidal anti-inflammatory drugs	6 (9.4)
Outcome	
Resolved	35 (54.7)
Improved	14 (21.9)
Ongoing pain at the end of the study	15 (23.4)

ROM, range of motion

pain might be due to incorrect shoulder exercises.[11,15] Some studies previously reported that an overhead pulley increased risk of shoulder pain because a traction force to the weak arm injures the soft tissue around the shoulder as the joint moves excessively in ROM.[3,16] This study confirmed that using an overhead pulley exercise was associated with shoulder pain significantly, and shoulder exercise with an overhead pulley should not be recommended in the post stroke patients.

This study showed 64.1% developed shoulder pain within a month, 17.2% within 2 months and 8 (12.5%) within 3 months after the onset of the stroke which was corresponding to the previous studies showing that most patients developed shoulder pain in the first month,[17,18] with an estimated prevalence of 17% in the first week and elevated throughout the recovery period with 20% to 24% the patients experienced shoulder pain from 1 to 16 months after stroke.[17] One large scale study revealed that self-reported shoulder pain increases from 256/1474 (17%) at one week and 284/1201 (23%) at six months.[18]

The major diagnoses of post stroke shoulder pain in this present study were impingement injury, adhesive capsulitis, central pain, complex regional pain syndrome, respectively. Most experts agreed that shoulder pain after stroke was not limited to a single pathology and affected by more than one pathologic condition.[5,18] Most patients would develop shoulder pain when they had muscle weakness and developed glenohumeral subluxation and scapular dyskinesis.[19] In the flaccid stage, the weight of the weak arm pulls down coracoacromial arch.[12] When patients raised the arm, soft tissue around the shoulder would squeeze the arch leading to impingement injury especially with an incorrect passive ROM exercise in forward flexion and abduction, like when using an overhead pulley. Another diagnosis of post stroke shoulder pain in this study was adhesive capsulitis which might be a result of immobilization and muscle wasting due to disuse. Adhesive capsulitis usually presents with pain and limitation in external rotation and abduction.[20] Painful stiffness of the shoulder adversely affects activities of daily living and consequently impairs quality of life.

For treatments of shoulder pain, physical modalities and medications were prescribed like in other studies.[6,11,21-23] The literature review also identified various treatment options to treat hemiplegic shoulder pain, such as electroacupuncture, therapeutic elastic taping, suprascapular nerve block and subacromial corticosteroid injection.[24-29] Although the patients in this study were treated with conventional treatments, the outcomes at the end of study was satisfactory with 76.6% resolved and improved, and only 13.4% still had ongoing shoulder pain that was similar to those admitted for inpatient rehabilitation in Thailand.[3] Early diagnosis and treatment may positively affect treatment outcomes.

This study had some limitations. It did not assess handling and transferring techniques which were reported to be associated with post stroke shoulder pain.[15] For further study, these factors should be assessed, collected and analyzed to confirm association with shoulder pain during home-based rehabilitation in post-acute phase.

In conclusion, post stroke shoulder pain was common during the first six months of post-acute home-based rehabilitation period. Early diagnosis and treatment could improve

or resolve shoulder pain. It was associated with impaired pinprick sensation, using an overhead pulley for shoulder exercise, and walking without a shoulder sling. An overhead pulley shoulder exercise should be abandoned in patients with stroke.

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Disclosure

The author has no conflict of interest to declare.

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Original Article

Food and Liquid Consistency Modification for Safe Swallowing in Elderly with Dysphagia Risk

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ABSTRACT

Objectives: This study aimed to evaluate the effect of food and liquid consistency modification on swallowing safety in healthy elderly with dysphagia risk.

Study design: Cross-sectional analytical study.

Setting: Outpatient clinic, Department of Rehabilitation Medicine, and Radiology Unit, Faculty of Medicine Ramathibodi Hospital, Mahidol University.

Subjects: Healthy elderly age > 65 who had mild to moderate dysphagia symptom, as defined by the 10-item Eating Assessment Tool (EAT-10) score ≥ 3

Methods: Each participant was performed a water swallowing test (WST), oropharyngeal physical examination, and videofluorographic swallowing study (VFSS). A bolus test, including four varying liquid consistencies and four modified food textures according to the International Dysphagia Diet Standardisation Initiative (IDDSI), was investigated. The Penetration-Aspiration Scale (PAS) was used to identify a primary outcome of safe swallow, which scores ≥ 2 was considered a high risk of penetration and aspiration. The residue in the oropharyngeal area was demonstrated by a pooling score (P-score), which was abnormal if ≥ 6. The pharyngeal transit duration (PTD) was also illustrated.

Results: Thirty-four subjects with a mean age of 72.0 (SD 6.8) years and mean EAT-10 score of 5.3 (SD 2.4) underwent VFSS. None of the subjects showed aspiration. The large volume of thin liquid (10 mL) revealed the highest frequency of penetration, 20.6%. There was no penetration during the test with 4 mL of moderately thick, extremely thick liquids, and all modified foods. A 10 mL of thin liquid and a pureed food were the highest occurrences of residue, 23.5%. Average PTD of regular food was the slowest at 120 ms, while small volume (4 mL) of thin liquid was the fastest at 69 ms.

Conclusion: In the elderly with dysphagia risk, a large bolus of thin liquid constituted the highest risk of penetration and aspiration. No penetration was found during the modified food test. However, a high frequency of abnormal pooling of residue was found after swallowing a pureed food and a large amount of liquid

Keywords: deglutition disorder, oropharyngeal dysphagia, food

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Introduction

Age-related change in swallowing mechanism (presbyphagia) is common among healthy senior citizens, with a prevalence of 40% in 65 years old or older.[1] There are many serious potential consequences of this condition, including malnutrition, dehydration, suffocation, aspiration, and aspiration pneumonia.[2,3]

There are several mechanisms by which presbyphagia can impair the swallowing process.[4,5] Regarding the oral phase of swallowing, tongue atrophy due to sarcopenia leads to a decrease in bolus propulsion force from oral to the pharyngeal cavity. Impaired oral sensation contributes to food sticking or pocketing in the oral cavity.[6] In some aging people, loss of tooth causes inadequate chewing of food. Age-related reduction of saliva production makes insufficient bolus forming, causing bolus transports to pharynx to be impaired.[7] In the pharyngeal phase, weakness of suprahyoid muscle and pharyngeal constrictor muscles cause bolus retention in vallecula and pyriform sinus area. Longer duration of swallowing results from decreasing pharyngeal sensation, the risk of laryngeal penetration and tracheal aspiration can be more pronounced.[5-7] Aspiration pneumonia also occurs commonly in those having aspiration.[4-8] To detect these symptoms of swallowing disorder, screening and self-assessment tests are popularly performed.

The common tests for swallow assessment include the water swallowing test (WST)[9] and the 10-items Eating

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Assessment Tool (EAT-10).[10] The WST is easily practiced as bedside screening, and the EAT-10 is frequently used to appraise the swallowing disorder by self-evaluation. However, these two techniques are used for screening of dysphagia risk. The videofluorographic swallowing study (VFSS) is a gold standard examination for detecting dysphagia and aspiration.[11,12] It also gives information on how to rehabilitate patients with dysphagia.

Food and liquid modification is a core component of swallowing rehabilitation in the elderly.[13] By changing a rate of eating and the physical properties of foods or liquids such as hardness, cohesiveness, and consistency, a bolus formation from oral to esophagus becomes more comfortable and safer. As a consequence, risks of bolus residue, choking, and suffocation decline. The International Dysphagia Diet Standardisation Initiative (IDDSI) has proposed standard terminology of food consistency and food texture modification.[14] A study from Catriona et al.[15] reported that thin liquid (IDDSI level 1) with low consistency and rapid flow rate increased the risk of laryngeal penetration, whereas mildly and moderately thick liquid (IDDSI level 2 and 3) with higher consistency and low flow rate decreased laryngeal penetration and aspiration. Homogenous consistency food (pureed food; IDDSI level 4) promotes comfortably chewing process, and reduces aspiration risk, as well as the study of Bingjie et al.[16] and Chen et al.[17] reported the same result.

There were many recommendations regarding food and liquid modification for patients with stroke and other neurological diseases. However, a study in the elderly with a subjective swallowing problem, but apparently healthy, was still lacking. Therefore, this study aimed to evaluate the effect of food and liquid texture modification of Thai foods in the aging population who had dysphagia risk.

Methods

Participants

We recruited volunteered participants from the outpatient Department of Rehabilitation Medicine, Faculty of Medicine, Ramathibodi Hospital, Mahidol University, during May 2017 – September 2017, with the following inclusion and exclusion criteria.

Inclusion criteria

- Age ≥ 65 years
- EAT-10 score ≥ 3
- Able to follow commands

Exclusion criteria

- Unable to communicate and answer the EAT-10
- Uncontrolled medical condition including unstable vital sign, and COPD or asthma exacerbation
- Allergy to contrast media
- History of disease or disorder that altered swallowing function such as stroke, Parkinson's disease, Alzheimer, motor neuron disease, multiple sclerosis, oropharyngeal anatomical distortion from head and neck cancer, and achalasia

Sample size calculation

After reviewing literature, the study of Kenichiro et al was selected for sample size calculation.[18] The study reported that the proportion of modified food and liquid penetration during the VFSS examination was 0.22. The power of 0.8 was set to determine the significant difference (5% type I error and 20% type II error). The sample size was calculated and a sample size of 34 participants was required.

Procedure

After obtaining informed consent, the EAT-10 (Thai version) questionnaire was completed by participants. For the illiterate subjects, one of the researchers read the questionnaire to the patients, asked them to answer, and filled in the form for them. Only those with the EAT-10 score of 3 or higher was defined as having dysphagia risk and the researcher proceeded to the next step.

Every subject received a dysphagia related physical examination and a water swallowing test (WST). The physical examination items included mouth opening, lip closure, tongue movement, and laryngeal excursion. For the WST, they swallowed 3 mL, 10 mL, and 50 mL of water. Change of respiratory rate ≥ 10% from baseline, choking, or wet voice after swallowing trials were considered abnormal and considered to fail the WST.[19] Next, they were scheduled for a VFSS.

The VFSS videos were acquired on a fluoroscopy (OmniDiagnost Eleva DI; Philips, Amsterdam, Netherland), carried out in lateral projection, at a frame rate 30 frames per second. The video was recorded by a digital video camera (Exmor R; Sony, Tokyo, Japan). The participants were comfortably sat in a wheelchair in an upright position. Different textures and consistencies of liquid and food mixed with contrast media were administered in order, as shown in Figure 1.

A fried rice was used as a regular diet sample, and as a base material for the preparation of other modified food consistency by the Institute of Nutrition, Mahidol University. All liquids were presented to the mouth of subjects in a 10 mL syringe. Pudding and foods were presented to the subjects in a regular tablespoon. Different consistencies of liquid and food texture modification were based on the IDDSI levels. An examiner gave a verbal cue for each participant to start each swallowing test. The VFSS was monitored live so that if aspiration were seen during the examination, the procedure would be stopped. Resuscitation and airway management were prepared ready for use within the examination room. If the residue was found at each bolus tested after first swallowing, the participant was instructed to repeat swallowing until there was no leftover bolus. In case of inability to clear the residue, transoral suction would be applied to empty the retention.

Outcome measurement

The demographic data were recorded. These include age, sex, body mass index (BMI), the EAT-10, and physical

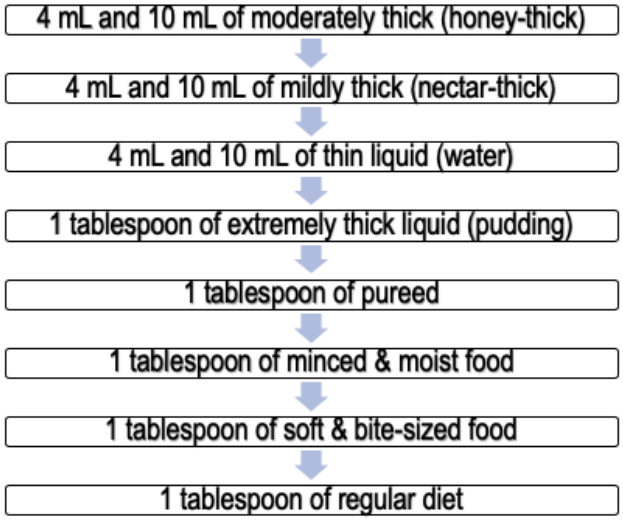

Figure 1. The order of liquid and food administration

examination findings. The VFSS video records were reviewed in a slow motion with frame-by-frame viewing when needed.

The severity of laryngeal penetration and aspiration of each swallowing conditions was assessed and scored separately according to the penetration aspiration scale (PAS) scoring criteria.[20,21] A score of 1 was given in case there was no observable penetration. Those with varying degrees of penetrations and aspirations received a score of 2 or greater. For the sake of this study, all of these were considered abnormal. The rating scale of PAS is shown as follows:

Score	Definition
1	No entry of bolus into larynx or trachea (no penetration or aspiration)
2	Penetration, entry of bolus into larynx above vocal cord and expelled out
3	Penetration, entry of bolus into larynx above vocal cord but
4	not expelled out
5	Penetration, bolus contacts vocal cord and expelled out Penetration, bolus contacts vocal cord but not expelled out
6	Aspiration, bolus enters the trachea and expelled out to larynx or pharynx
7	Aspiration, bolus enters the trachea but not expelled out following attempts
8	Aspiration, bolus enters the trachea without attempts to expel

The severity of bolus retention in the larynx was classified according to the pooling score (P-score).[22] It comprises of 3 domains: location, amount of residue, and the number of repeated swallowing needed to clear residue. The P-score of 6 and higher were considered abnormal. The P-score definition was illustrated as follows:

The pharyngeal transit duration (PTD) was recorded as the time from when the bolus head pass the ramus of mandible, to the time when the tail of the bolus has passed into the upper esophageal sphincter (UES). All scoring and timing were measured by one of the authors (BK) and reviewed by another (PB). If conflicting scores were found,

P-score		Score
Location of residue	Valleculae	1
	Pyriform sinus	2
	Larynx/vocal cord	3
	Below larynx	4
Amount	Coating	1
	Minimum	2
	Maximum	3
Management to clear the residue (repeated swallow)	< 2	2
	2 – 5	3
	> 5	4
Total score	No dysphagia	4-5
	Mild dysphagia	6-7
	Moderate dysphagia	8-9
	Severe dysphagia	10-11

the video recordings and the data forms were reviewed until an agreement was reached.

Statistical analyses

The STATA version 15 (Stata Corp LLC, Texas, USA) was used to analyze the result. The Shapilo-Wilk test was used to evaluate the normality of data distribution. The demographic data were reported as frequency and mean (SD). The PAS score and P-score were shown in the frequency of abnormality, which was PAS ≥ 2 and P-score ≥ 6. The PTD of each liquid and food type were compared by using ANOVA. The $p < 0.05$ was considered to be a statistical significance.

This study was approved by the Committee on Human Rights Related to Research Involving Human Subjects, Faculty of Medicine Ramathibodi Hospital, approval number ID12-59-02.

Results

Table 1 shows the demographic data of 34 participants (19 females and 15 males) enrolled in this study. The mean age was 72.0 (SD 6.8) years old. The EAT-10 score was 5.8 (SD 2.4), which was mild to moderate dysphagia risk. Thirty-two participants passed all WSTs. Only two participants had a post-swallowing cough when tested with 50 mL of water. The PE findings were normal in all participants.

Modified liquid swallowing test (Table 2)

The first and second-largest percentage of subjects with a "failed" PAS score of 2 or greater, were found in the case of large volume 10 mL of thin liquid test (20.6%) and the case of small volume 4 mL of thin liquid (14.7%). 5.9% of the subjects showed abnormal PAS when swallowing 10 mL and 4 mL of thick liquid. Only one subject (2.9%) demonstrated abnormal PAS during the test with 10 mL of moderately thick liquid. The rest of the liquid swallowing tests were free from penetration.

Some subjects showed abnormal pooling, as defined by P-score of 6 or higher, in every liquid bolus condition tested. VFSS test with 10 mL thin liquid, resulted in the highest rate

Table 1. Demographic data of 34 participants

Data	Results
Sex, Male[1]	15 (44.0)
Age[2]	72 (7)
BMI2 (kg/m^2)	25.8 (4.9)
EAT-10[2]	5.8 (2.4)
Physical examination[1]	
- Normal mouth opening	34 (100.0)
- Normal lip closure	34 (100.0)
- Normal tongue movement	34 (100.0)
- Normal Laryngeal elevation	34 (100.0)
WST (passed the screening test)[1]	
- 3 mL (MWST)	34 (100.0)
- 10 mL	34 (100.0)
- 50 mL	32 (94.1)

[1]Mean (SD), [2]number (%)

BMI, body mass index; WST, water swallowing test; MWST, modified water swallowing test

of abnormal P-score of 23.5%, followed by 10 mL of mildly thick liquid (nectar) and 10 mL of moderately thick liquid, 17.7%. When compared in the same type of liquid, a more massive amount (10 mL) had a more abnormal P-score than a lesser amount (4 mL).

The slowest and the fastest PTD among the liquid swallowing test were the pudding-like (extremely thick liquid) at 97 msec, and the 4 mL-thin liquid at 69 milliseconds. There was an observable trend showing faster PTD in smaller volume and thinner consistency liquid than in larger volume and thicker consistency. There was a significant difference in PTD between 10 mL thin liquid, 71 (SD 23) ms, and extremely thick liquid, 97 (SD 33) ms, *p*-value = 0.003, 95%CI 13-38.

Modified food swallowing test (Table 2)

There was no penetration and aspiration seen during VFFS while testing with any type of modified food. The average P-score during thick pureed food swallowing condition was the highest among all the food swallowing tests at 23.5%, followed by minced and moist (20.6%), and regular diet (17.7%). As regarding PTD, swallowing regular food was the most prolonged (120 msec). However, there was no significant difference of PTD among different food types

Discussion

The enrolled healthy elderly had a mean score of the EAT-10 score, 5.8 (SD 2.4), a mild subjective symptom. However, the WST showed that two participants choked with 50 mL of thin liquid. Moreover, a higher frequency of abnormal penetration and pooling was reported when swallowing a larger amount in each type of liquid bolus during VFSS. These results are compatible with previous studies that in healthy elderly, the penetration and retention rate increased with higher volume of liquid.[23,24] Therefore, a large amount of liquid poses the highest risk.

For the VFSS findings with modified liquids, this study showed that thicken liquid (IDDSI 2-4) had lesser penetration and aspiration frequency than thin liquid. The more massive amount of liquid, the more occurrence of penetration and aspiration, was indicated. The 10 mL of thin liquid (IDDSI 0) established the highest rate of PAS ≥ 2, followed by 4 mL of thin liquid. No participants showed unsafe swallowing during testing with extremely thick liquid (pudding) and 4 mL of moderately thick (honey-thick) liquid. The thicken liquid had more prevalence of pharyngeal residue. The higher viscosity, the higher rate of residue was noted. The study of Catriona et al. also reported the same result that was the higher viscosity and lower velocity decreased the risk of material enter into the larynx and below vocal cord but increased the risk of post-swallowing residue in the pharynx, especially in the elderly who had dysphagia risk.[15] The thin liquid which had the lowest viscosity was the top of penetration and aspiration rate. Troche et al. displayed that pudding thick (extremely thick liquid) had a lower PAS score than thin liquid.[25] As the results from our study, we would say that older people

Table 2. The number of abnormality of PAS (PAS > 2), and P-score (P-score > 6) in each type of liquid and food modification

Material	PAS ≥ 2[1] N=34	P-score ≥ 6[1] N=34	PTD (ms)[2]
Modified liquid			
Thin liquid 4 ml (IDDSI 0)	5 (14.7)	3 (8.8)	69 (22)
Thin liquid 10 ml (IDDSI 0)	7 (20.6)	8 (23.5)	71 (23)
Mildly thick liquid 4 ml (IDDSI 2)	2 (5.9)	5 (14.7)	79 (24)
Mildly thick liquid 10 ml (IDDSI 2)	2 (5.9)	6 (17.7)	81 (26)
Moderately thick liquid 4 ml (IDDSI 3)	0 (0.0)	3 (8.8)	80 (21)
Moderately thick liquid 10 ml (IDDSI 3)	1 (2.9)	6 (17.7)	84 (23)
Extremely thick liquid (IDDSI 4)	0 (0.0)	2 (5.9)	97 (33)
Modified food			
Pureed (IDDSI 4)	0 (0.0)	8 (23.5)	119 (46)
Minced & moist food (IDDSI 5)	0 (0.0)	7 (20.6)	107 (27)
Soft & bite-sized food (IDDSI 6)	0 (0.0)	4 (11.8)	101 (40)
Regular food (IDDSI 7)	0 (0.0)	6 (17.7)	120 (45)

[1]Number (%), [2]mean (SD)

PAS, penetration-aspiration scale; P-score, pooling score; PTD, pharyngeal transit duration; IDDSI, International Dysphagia Diet Standardisation Initiative

should consume a small amount of clear water. Even though thicker liquids resulted in less pharyngeal residue after swallowing, the retention percentage was between 8.8%-17.7%. Repeated dry swallowing and throat clearing after ingestion of thickening liquids and frequent small sip rather than taking a big gulp of liquid should be recommended to this risk group.

Despite the reassuring finding that the rate of penetration during food swallowing was zero, it was worth mentioning that the rate of retention was very high in the case of minced & moist (IDDSI 5) (20.6%) and pureed (IDDSI 4) (23.5%). If swallowing of such food is repeated, the amount of retained food in the larynx may accumulate. This retention could consequently lead to post-swallowing aspiration. For this reason, we should not recommend pureed food over regular food for this group of people. The properties of a safest food with low retention is high adhesiveness, low cohesiveness, and low hardiness. The pureed and minced & moist food should match the most secured texture.[18,26] It was a contrast to the current result. A future study that considers "stickiness" of food bolus and swallowing risk would be needed to guide reasonable recommendations on food modification for this population. With the available data at hand, it seems that pudding-like or perhaps jelly-like food with low adhesion to the pharyngeal wall and a slow transit time may be the best suggestion for the healthy elderly population with mild to moderate dysphagia symptom to avoid aspiration and penetration.

This study found that the higher viscosity, the longer pharyngeal transit duration (PTD) was needed. The study of Catriona also demonstrated the same result that was the most prolonged PTD was found in an extremely thick liquid.[15] Slowing the flow of liquid allows more time for the airway protection mechanism. Thereby, the more thickening of liquid, the less risky laryngeal penetration and tracheal aspiration.[27-29] We would recommend that in the elderly, even with mild swallowing problems, slightly more viscosity would reduce the complication.

The limitation of this study was that each bolus consistency was tested only once. Therefore, it was plausible to expect that repeated swallowing, in a situation with distracting environmental factors, could result in a higher penetration rate. It is possible that in real life situation aspiration and choking can happen. This issue would be in a future study.

We can conclude that in the healthy elderly with dysphagia risk, ingesting a larger volume (10 mL) of thin liquid related to the highest risk of penetration and aspiration. A small amount of moderately thick liquid (4 mL) was the safest. In mild dysphagia cases, food types did not affect penetration and aspiration but high viscosity and cohesive food increased the residue rate in the pharyngeal area. Hence, soft & bite-sized food was recommended for this population to avoid food retention.

Disclosure

All authors declare no conflict of interest of any kind.

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Original Article

Efficacy of Sedentary Time Reduction with Mobile Texting and Focused Educational Sessions in Patients with Coronary Artery Disease: a Randomized Controlled Trial

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ABSTRACT

Objectives: To assess the efficacy of mobile texting and focused educational session on sedentary behavior in patients suffering from coronary artery disease.
Study design: Randomized controlled trial.
Setting: Cardiac Rehabilitation Clinic, Rehabilitation Medicine Department, Faculty of Medicine, Ramathibodi Hospital, Mahidol University, Bangkok, Thailand.
Subjects: Patients with coronary artery disease who underwent cardiac rehabilitation program from November 2018 to May 2020 at Ramathibodi Cardiac Rehabilitation Clinic.
Methods: The participants were randomized to receive either a standard cardiac rehabilitation (control group), or a standard cardiac rehabilitation with mobile texting and focused educational sessions on sedentary behavior (intervention group). Sedentary time and time spent in other physical activity levels were recorded at baseline, 1, 3, and 6 months following participation in both groups. This was done using the Thai short International Physical Activity Questionnaire (Thai short IPAQ). The effects of mobile texting and focused educational sessions on sedentary behavior were analyzed by employing a mixed effects linear regression analysis.
Results: Participants spent about 13.72 hours/day engaging in sedentary behavior. The intervention group showed significantly less prevalence of diabetes compared with the control group. This baseline difference was adjusted in the regression model. The between-group comparison study found that sedentary time was significantly less at 3 months in the intervention group compared with the control group (at 3 months; difference 1.75 hour/day, p = 0.002). Mild intensity physical activity rose significantly only at the end of the study (at 6 months; difference 1.00 hour/day, p = 0.047). There was no significant difference between the two groups at all follow-up intervals for moderate-to-vigorous physical activity.
Conclusion: Additional mobile texting and focused educational sessions on sedentary behavior to a standard cardiac rehabilitation program, helped the patients with coronary artery disease reduce their sedentary time at 3 months.

Keywords: behavior, patient education, cardiac rehabilitation, physical activity, coronary artery disease

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Introduction

Sedentary behavior is described as waking behaviors consuming energy expenditure amounting to less than 1.5 metabolic equivalents (METs).[1] It comprises sitting or lying down with or without performing other activities that also entail minimal energy expenditure such as using a computer, mobile phone, or watching television. Sedentary behavior is closely linked to increased risk of cardiovascular disease, diabetes and metabolic syndrome.[1] There is no generally accepted guideline for the proper amount of time spent in a prolonged sitting, but a previous study revealed that all-cause mortality increased by about 5% for every one hour spent sitting for longer than 7 hours per day.[2]

Physical activity is any bodily movement that increases energy expenditure above the basal metabolic rate (1 MET). Sedentary behavior comprises physical activities with energy expenditure ranging from 1 to 1.5 METs. Light physical activity is defined as activities with energy expenditure of 1.6 to 2.9 METs. Moderate-to-vigorous physical activity refers to activities which require energy expenditure of 3 METs or greater.[3] Recommendations for the physical activity level in order to maintain good health include at least 150 minutes a week of moderate intensity or 75 minutes a week of vigorous intensity aerobic physical activity.[3]

Interestingly, sedentary behavior and a lack of moderate-to-vigorous physical activity should be considered as separate risk factors because they affect mortality risk independently.[1] This is because sedentary individuals who are unable to meet exercise recommendations seem to be at a high risk of mortality rate when compared to individuals with either risk factors.[1] For this reason, sedentary behavior is

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one of the modifiable risk factors for patients suffering from coronary artery disease (CAD). However, the standard cardiac rehabilitation (CR) usually focuses on increasing moderate-to-vigorous physical activity, but often fails to reduce patients' sedentary time.[4-7] Furthermore, patients and healthcare providers in cardiac rehabilitation clinics are usually concerned with increasing physical activity more than reducing sedentary time.[8] Another study reported that patients with heart disease were prone to experiencing more sedentary time compared with those without heart disease in the same age group.[9]

To evaluate physical activity, two very commonly used measurements, including self-report questionnaires (subjective method) and physical activity monitors by accelerometers (objective method), are described.[3] Between these two methods, the questionnaires are inexpensive, require less time to administer when compared to the physical activity monitors, and can access various types of physical activity (i.e. gardening, walking, etc.).[3] One standard questionnaire for describing physical activity is the International Physical Activity Questionnaire (IPAQ), which has been translated into the Thai language.[10] The validity and reliability of the Thai version of IPAQ (Thai short IPAQ) were acceptable when compared with physical activity monitors by accelerometers.[10]

Many interventions have been proposed in order to decrease sedentary behavior, including behavioral therapy. Previous studies found that behavioral therapy can significantly improve sedentary time.[9] However, this process requires specialized personnel such as behavioral therapists, who are quite rare in developing countries such as Thailand. On the other hand, text message intervention is another intervention previously reported as able to produce behavioral change.[11,12] According to the self-determination theory, which is one of the motivational theories commonly referred to in the field of health promotion, mobile texting can help provide recommendations for rationales, goals, and choices about sedentary time reduction.[13] These could be indicated as an outside source of extrinsic motivation for encouraging patients to curtail their sedentary time. Meta-analyses have demonstrated the beneficial effects of text message intervention on health behavioral change.[11,12,14,15] This method is convenient, low cost, and does not require healthcare providers with sophisticated skills.

Nevertheless, we though that text message intervention alone can provide only one-way communication since there is no feedback facility. Combined with focused educational sessions, this method could provide two-way communication which allows more patients to participate; also, one previous meta-analysis study showed that supplemented text messages with additional components including face-to-face consultations are more effective in promoting behavioral change.[11] However, another analysis exploring the effects of texting intervention in patients with CAD offered information on multiple factors for secondary prevention, but was not focused on sedentary behavior and physical activity.[12] To the best of our knowledge, no study has yet investigated the effects of combined text message intervention and focused educational sessions on sedentary behavior and physical activity.

The primary objective of the study was to evaluate the effects of mobile texting and focused educational sessions on sedentary behavior in patients with CAD who received a standard CR. We hypothesized that patients who additionally received both mobile texting and focused educational interventions could further reduce their sedentary time when compared with those who received a standard CR alone.

Methods

Ethical approval was obtained from the Ramathibodi Hospital Human Research Committee (approval ethical number/RF_62026). Patients were given information and documented their informed consent.

Participants

They were patients with CAD who received optimal treatment, visited an out-patient cardiac rehabilitation clinic of Ramathibodi Hospital from November, 2018 to May, 2020, and provided an informed consent to participate this study.

Inclusion criteria

- Stable CAD
- More than 7 hours of sedentary time per day
- Being able to read Thai
- Having a personal mobile phone with Line application (a freeware application for instant communications on electronic devices) or having other persons to receive the messages and communicate their content to them

Exclusion criteria

- Contraindication for exercise training, severe obesity, pregnancy, severe chronic obstructive pulmonary disease, orthopedics, neurological or peripheral vascular diseases that may compromise safety in improving physical activity

Interventions

The study had two interventions which were the text messages and the focused educational sessions on sedentary behavior.

1. The text messages were developed by the researchers. The content of the messages was adapted from guidelines or recommendations for CR,[3] exercise prescription,[16] and interventions for reducing sedentary behavior.[17-21]

The development of text messages involved an expert panel which included one physiatrist, one nurse who worked in the cardiac rehabilitation clinic, two physical therapists who worked in the cardiac rehabilitation clinic, one social worker, and two persons who were not medical providers. Three criteria were used in the evaluation: 1) usefulness of the messages in reducing sedentary time; 2) readability; and 3) a final open question for feedback and any suggestions. The stated messages should be approved by at least 5/7

experts for both usefulness and readability purposes. Unaccepted messages were changed in accordance with the expert group's suggestions.

2. The focused educational sessions on sedentary behavior comprised 4 sessions. The physical therapists gave information about each session to participants. Each physical therapist gave the information in the same standard of content at baseline, 1, 3, and 6 months, respectively.

The first session was concerned with the definition of sedentary behavior, physical activity and impact of sedentary behavior on health. Furthermore, the participants were encouraged to set goals to reduce sedentary time. The second session involved information regarding SMART goals (Specific, Measurable, Attainable, Realistic, and Time-Bound), and a proper environment to reduce sedentary time. In the third session, participants were informed about techniques to reduce sedentary time and how to adjust their goals appropriately. For the last session, participants reviewed all the information from the previous 3 sessions and summarized their long-term home-program with the physical therapist.

To reduce the problem of bias in this study, the physical therapists who provided focused educational sessions on sedentary behavior were not the same persons who conducted the standard CR.

Measurements

Physical activity was assessed subjectively using the Thai short International Physical Activity Questionnaire (Thai short IPAQ).[10] The Thai short IPAQ is a questionnaire designed to estimate time employed in different physical activity levels (sedentary, mild intensity, intensity, vigorous intensity) in the past week (hours and minutes). The Thai short IPAQ was validated in Thailand and demonstrated a fair correlation with accelerometers.[10] The primary outcome of this study was sedentary behavior. Other physical activity levels were the secondary outcomes.

Sample size calculation

The sample size calculation was based on the primary outcome, which was a reduction in sedentary time, according to the study by Thakkar J, et al (2016),[12] assuming an alpha level (α) of 0.05, and desired power of 80%.

The estimated sample size was calculated to be 33 individuals per group. Allowing for a 20% dropout rate, there were 40 subjects per group.

Procedures

The experiment was a randomized controlled trial study. Randomization was 1:1 ratio with a block of four. Allocation was concealed from physical activity assessors and data analysts. We recorded baseline characteristics including age, sex, nationality, BMI, level of education, current employment status, marital status, smoking status, treatment, underlying disease, risk of exercise, and sitting time (hours/day).

Participants in the intervention arm received the mobile texting, the focused educational sessions on sedentary behavior and the standard CR. The control arm received only the standard CR. The messages (1 message/day, 4 days/week, on random days, between 08:00 h and 16:00 h) offered information on definitions of physical activity and sedentary behavior, sedentary behavior health links, general encouragement, instructions, goal setting, self-monitoring, social support, etc. The messages were sent for 6 months.

The standard cardiac program consisted of patient assessment, risk stratification, detection of depressive symptoms, and educational sessions on physical activity which did not focus on sedentary behavior, nutritional status, and secondary prevention. The program took place at the 1st visit, and approximately at 1-month, 3-month, and 6-month intervals. Physical activity data assessment was conducted at the CR clinic where the CR program took place or via telephone by 2 health professionals of the same standard.

Sedentary time and time spent on other physical activity levels were assessed at baseline, 1, 3, and 6 months after having participated in the study.

Statistical analysis

We used STATA version 14. Descriptive data were reported using mean, standard deviation, frequency, and percentage. The modified intention-to-treat was used to analyze the data. If participants were lost to follow-up at all intervals, they would be considered as dropouts and excluded from the analysis. However, if they were lost to follow-up at some intervals, any collected data would be used in the analysis. To analyze the effects of the mobile texting and the focused educational sessions on sedentary behavior, mixed effects linear regression analysis was employed.

Results

One hundred participants were randomized in the study from November 2018 to May 2020. The dropouts included five participants in the control group and nine participants in the intervention group (Figure 1). Most participants were lost to follow-up due to medical and transportation problems. One patient in the intervention group died from an underlying disease while sleeping and was not associated with increased physical activity.

Most patients were males, working, married, never smoked, received CABG surgery, had hypertension, and had low risk of exercise (Table 1). On average, all participants reported sitting time, light physical activity, and moderate-to-vigorous physical activity to be about 13.72 hours/day, 1.16 hours/day, and 0.03 hours/day, respectively. The intervention group showed a significantly lower prevalence of diabetes than the control group. This baseline difference was adjusted in the regression model.

In the intra-group comparison study with the baseline (Table 2), both study arms revealed a significant decrease

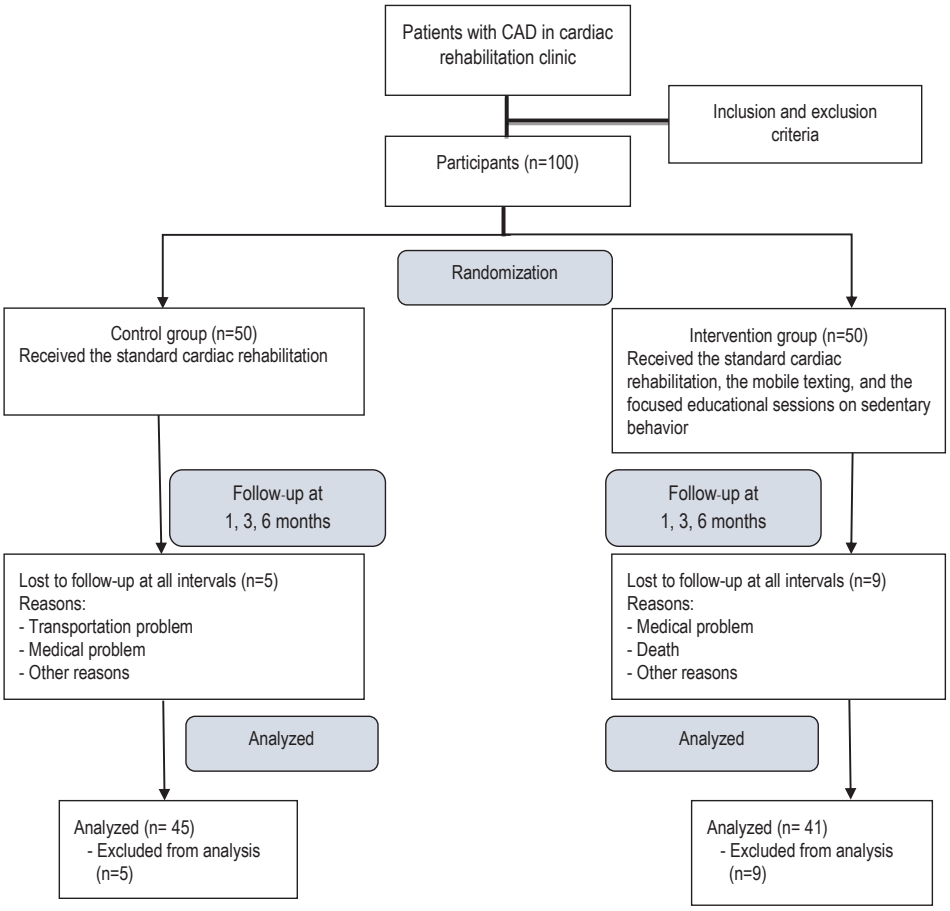

Figure 1. Flow chart of subjects' participation

in sedentary time. In the control group, sedentary time significantly dropped at 1 month (difference -1.18 hour/day, p = 0.003), 3 months (difference -1.00 hour/day, p = 0.017), and 6 months (difference -1.13 hour/day, p = 0.007). In the intervention group, sedentary time significantly decreased at 1 month (difference -1.32 hour/day, p = 0.002), 3 months (difference -2.36 hour/day, p = 0.000), and 6 months (difference -1.39 hour/day, p = 0.002).

Time spent in light physical activity significantly increased at all follow-up intervals for both groups. In the control group, light physical activity significantly increased at 1 month (difference 0.91 hour/day, p = 0.014), 3 months (difference 1.19 hour/day, p = 0.002), and 6 months (difference 0.92 hour/day, p = 0.018). For the intervention group, light physical activity significantly increased at 1 month (difference 1.14 hour/day, p = 0.005), 3 months (difference 1.56 hour/day, p = 0.000), and 6 months (difference 1.82 hour/day, p = 0.000). There were no significantly different changes in time spent on moderate-to-vigorous physical activity at baseline, and any follow–up intervals.

For the between-group comparison study (Table 2), sedentary time was significantly lower only at 3 months for the intervention group compared with the control group (difference -1.75 hour/day, p = 0.002). Light physical activity for the intervention group significantly increased only at the end of the study (difference 1.00 hour/day, p = 0.047) and there was no significant difference between the two groups at all follow-up intervals for moderate-to-vigorous physical activity.

Discussion

According to previous studies from Canada and Portugal conducted by Biwas et al.[4] and F.Ribeiro et al.[6] the mean sedentary time at baseline was 8.1 hours/day and 6.47 hours/day respectively. In our study, the mean sedentary time amounted to 13.72 hours/day. Our findings show that baseline sedentary time in patients with CAD seems to be higher than in the previous studies. The difference in baseline sedentary time compared to the previous analyses may be due to our inclusion criteria which state that the patients should have a sedentary time lasting more than 7 hours per day. There is no generally accepted guideline for significantly prolonged sedentary time. One previous meta-analysis study shows every one hour spent in a sedentary state for longer than 7 hours per day means an increase in all-cause mortality by about 5%.[2] So, we used these criteria for selecting participants who had a risk factor of too much sedentary time.

The change in sedentary time during CR in the intra-group study lasted for 6 months for both groups which corresponds with the study by Nienke, et al.[5] which shows that CR had long-term effects in reducing sedentary time. However, the improvement differed from the studies of Biwas, et al.[4] and

Table 1. General characteristics of patients in the intervention and the control groups

Characteristics	Control group (n=45)	Intervention group (n=41)
Age[1]	62.96 (7.41)	64.63 (9.32)
Male[2]	34 (75.56)	26 (63.41)
Female[2]	11 (24.44)	15 (36.59)
Thai[2]	44 (97.78)	41 (100)
BMI[1]	25.01 (4.00)	24.43 (4.07)
Level of education[2]		
None/primary	15 (34.09)	9 (23.08)
Secondary	9 (20.45)	9 (23.08)
Vocational training	8 (18.18)	9 (23.08)
University	12 (27.27)	12 (30.77)
Current employment status[2]		
Working	20 (46.51)	15 (42.86)
Not working	12 (27.91)	9 (25.71)
Retired	11 (25.58)	11 (31.43)
Marital status[2]		
Unmarried	3 (6.82)	5 (12.82)
Married	36 (81.82)	32 (82.05)
Divorced	2 (4.55)	1 (2.56)
Separated	0	0
Widowed	3 (6.82)	1 (2.56)
Smoking status[2]		
Current smoker	0	0
Ex-smoker	20 (45.45)	14 (35)
Never smoked	24 (54.55)	26 (65)
Treatment[2]		
CABG	45 (100)	40 (97.56)
PCI	0	1 (2.44)
Thrombolysis	0	0
Underlying disease[2]		
Diabetes	24 (53.33)	12 (29.27)
Hypertension	38 (84.44)	31 (75.61)
COPD	1 (2.22)	2 (4.88)
CKD	8 (17.78)	11 (26.83)
Risk of exercise[2]		
Low risk	39 (86.67)	37 (90.24)
Intermediate risk	3 (6.67)	2 (4.88)
High risk	3 (6.67)	2 (4.88)
Physical activity from Thai short IPAQ (hour/day)[1]		
Sedentary time	13.90 (1.98)	13.53 (2.14)
Light physical activity	1.12 (1.46)	1.22 (1.55)
MVPA	0.04 (0.15)	0.02 (0.06)

[1]Mean (SD), [2]number (%)

BMI, body mass index; CABG, coronary artery bypass graft; PCI, percutaneous coronary intervention; COPD, chronic obstructive pulmonary disease; CKD, chronic kidney disease; IPAQ, the international physical activity questionnaire; MVPA, moderate-to-vigorous physical activity

Ribeiro, et al,[6] both of which show the standard CR seems to fail in changing sedentary behavior. Consequently, the effect of standard CR in reducing sedentary time was unclear. Due to the lower baseline sedentary time of the previous studies,[4,6] the improvement of sedentary time for both groups in our study may be from the ceiling effect of standard CR. We assume that general standard CR may have the ceiling to reduce sedentary time. Thus, other behavioral interventions such as the mobile texting and the focused educational sessions on sedentary behavior should be added to boost the effectiveness of standard CR on sedentary time reduction.

For the between-group study, the intervention group reported a greater decrease in sedentary time than the control

Table 2. Changes in sedentary time, light physical activity, and moderate-to-vigorous physical activity from baseline to 1-month, to 3-month, and to 6-month follow-up

	1-month Coef. (95%CI)	*p*-value	3-month Coef. (95%CI)	*p*-value	6-month Coef. (95%CI)	*p*-value
Sedentary time (h/day)						
Control group	-1.18 (-1.96- -0.40)	0.003*	-1.00 (-1.82- -0.18)	0.017*	-1.13 (-1.95- -0.31)	0.007*
Intervention group	-1.32 (-2.15- -0.49)	0.002*	-2.36 (-3.20- -1.53)	0.000*	-1.39 (-2.24- -0.53)	0.002*
Between-group	-0.53 (-1.61-0.55)	0.339	-1.75 (-2.87- -0.64)	0.002*	-0.65 (-1.78- 0.48)	0.261
Light physical activity (h/day)						
Control group	0.91 (0.18-1.63)	0.014*	1.19 (0.43-1.95)	0.002*	0.92 (0.15-1.68)	0.018*
Intervention group	1.14 (0.34-1.93)	0.005*	1.56 (0.77-2.35)	0.000*	1.82 (1.02-2.62)	0.000*
Between-group	0.32 (-0.63-1.27)	0.505	0.46 (-0.52-1.43)	0.357	1.00 (0.01-1.98)	0.047*
Moderate-to-vigorous physical activity (h/day)						
Control group	0.08 (-0.05-0.20)	0.218	0.02 (-0.11-0.14)	0.803	0.04 (-0.09-0.16)	0.585
Intervention group	-0.00 (-0.14-0.13)	0.957	0.11 (-0.02-0.24)	0.091	0.11 (-0.02-0.25)	0.103
Between-group	-0.11 (-0.25-0.03)	0.127	0.07 (-0.07-0.21)	0.343	0.05 (-0.10-0.19)	0.515

*Statistically significant, $p < 0.05$

group in the 3rd month. This suggests that the information from the mobile texting and the focused educational sessions on sedentary behavior helped participants in the intervention group to become less sedentary, but without significant difference in the long-term follow-up when compared with the control group. This may be due to the rare attainment of behavioral change maintenance,[22] but there was a minimal reduction in sedentary time in the intervention group at the 6–month interval. In the future, this intervention pattern needs to be evaluated to maintain sedentary time reduction in the long-term period.

Previous studies reported an improvement in moderate-to-vigorous physical activity after CR. Nevertheless, the increase was not detected in our study. This may be because our participants were more sedentary than the patients who participated in CR in other analyses, so they decided to do light physical activity, instead of moderate-to-vigorous physical activity. Mild intensity physical activity in both control and intervention groups seemed to improve following CR and significantly improved for the intervention group compared to the control group at the end of the study. In addition, the CR program for severely stationary patients of our institute is designed to start with low-intensity exercise and to have slow progression. Therefore, our CR protocol may influence an improvement in moderate-to-vigorous physical activity. This outcome indicates that after CR, moderate-to-vigorous physical activity remains the same. The American Association of Cardiovascular and Pulmonary Rehabilitation (AACVPR) recommends 150 minutes of moderate physical activity per week,[3] but no participants attained that level.

Increase in moderate-to-vigorous physical activity and reduction in sedentary time are not the same, given that the standard CR prefers less focus on reducing sedentary time to increased exercise. For severely inactive cardiac patients, we suggest assessing sedentary time as a risk factor and monitoring it as an outcome of CR program because the patients tend to change sedentary time to light physical activity instead of increasing moderate-to-vigorous physical activity. Moreover, concentrating on reducing sedentary behavior helps reduce sedentary time greatly compared with the standard CR.

Our study was limited by the lack of access to the maintenance of sedentary behavior reduction. Long-term follow-up was not well assessed to evaluate whether the sedentary time and the physical activity decline after the completion of CR had been achieved. This is because there were studies that did find that patients within a year of CR completion were as sedentary as new CR patients.[23] Future studies should undertake long-term follow-ups to answer this question. The study did not evaluate satisfaction or dissatisfaction of the intervention. Moreover, it did not evaluate adherence of the participants in the intervention group, or whether they read or understood the messages or not. In addition, the follow-up period of some participants was not completed and there were some missing data during the study. We excluded some participants who were lost to follow-up at all intervals from our statistical analysis because we could not infer the trend of their sedentary behavior, light physical activity, and moderate-to-vigorous physical activity. Therefore, our study did not conduct a general intention-to-treat analysis.

In conclusion, mobile texting and focused educational sessions on sedentary behavior helped patients with coronary artery disease reduce sedentary time at the 3-month interval and additionally increased light intensity physical activity at 6-month interval in patients who received the standard CR. These methods can be used complementarily to the standard CR due to its efficacy, convenience, and low cost.

Disclosure

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Original Article

Negative Effects of Betahistine on Recovery of Brainstem Oculo-motor Integration after Vestibular Rehabilitation of Benign Paroxysmal Positional Vertigo (BPPV) Patients

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ABSTRACT

Objectives: To evaluate effects of betahistine, an antihistamine, on recovery of vestibulo-ocular functions in patients with BPPV after vestibular rehabilitation (VR) therapy.
Study design: Retrospective study.
Setting: Vestibular Rehabilitation Clinic, Division of Physical Therapy, Department of Rehabilitation Medicine, Trang Hospital, Ministry of Public Health, Trang, Thailand.
Subjects: Patients with BPPV who were referred for VR; assessed with global BPPV symptom severity visual analog scale (VAS), Dix-Hallpike test (DHT), roll test (RT), head thrust test (HTT), and gaze evoked nystagmus test (GENT) before once a week of VR and one week after completing three sessions; and performed a daily home-based VR exercises (VREs) for 3 or 4 weeks.
Methods: Data of all assessments mentioned above were extracted from case record forms, and divided into two groups: those taking betahistine (81 patients) and those not taking any antihistamine (84 patients). Data from the two groups were compared and analyzed.
Results: After completing all three sessions of VR therapy, every assessment score significantly decreased ($p < 0.001$) in both groups. Before the first therapy, mean VAS scores (SD) of the betahistine and the no antihistamine groups were 9.12 (0.73) and 9.22 (0.70), respectively ($p = 0.38$); in the second assessment, were 4.17 (0.86) and 5.15 (1.21) respectively ($p < 0.001$); in the third assessment, were 3.53 (0.63) and 2.57 (3.32) ($p < 0.001$), and in the last assessment, were 1.84 (0.64) and 0.03 (0.18) respectively ($p < 0.001$). Regarding the baseline assessment of the DHT, the RT, the GENT, and the HTT, there were no significant differences ($p > 0.01$) between the two groups. However, in all subsequent assessments there were significant differences in the GENT and the HTT scores between the two groups, favoring to the no antihistamine group over the betahistine group ($p < 0.01$). The DHT and the RT scores did not reach significant differences between the two groups in the last two weeks of assessments.
Conclusion: Once a week of VR therapy and a daily home-based VREs for three or four weeks significantly decreased the BPPV symptoms. Recovery of vestibulo-ocular reflex function seemed less and not as complete in those taking betahistine.

Keywords: vestibular rehabilitation, physical therapy, benign paroxysmal positional vertigo, vestibulo-ocular reflex, betahistine

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Introduction

Benign paroxysmal positional vertigo (BPPV) is the most common cause of peripheral vertigo.[1] It is caused by displaced calcium carbonate particles called otoliths (or otoconia) inside the semicircular canals of the vestibular labyrinth of the inner ear.[2] Factors found to be correlated with increased risk of having BPPV attack are the following: elderly age,[3] vestibular artery flow impairment,[4] and cardiovascular risk factors such as diabetes, dyslipidemia, hypertension, etc.[5] Mechanical shock such as that produced during dental surgery could possibly be a precipitating cause.[6]

BPPV is clinically diagnosed by observing nystagmus and subjective vertigo during the so-called BPPV provocation tests such as Dix-Hallpike test (DHT), head thrust test (HTT), etc.[7] Each of these tests mobilizes the otoliths in one of the three semicircular canals, through a specific head movement. It is important to rule out serious diseases which mimic symptoms of BPPV such as stroke, transient ischemic attack, and posterior fossa brain pathology.[8]

Impact of BPPV ranges from mild annoyance to highly debilitating. It affects safety and falling risk. Two most commonly recommended rehabilitation methods are canalith repositioning procedure (CRP)[9] and vestibular rehabilitation exercises (VREs). Combination of CRP and VREs are expected to be more effective that either one alone,[10] especially in the long-term reduction of BPPV severity scores.[11]

Histamine receptor antagonists are the most commonly prescribed medication for BPPV,[12] but the mechanisms which this group of medication alleviates BPPV related symptoms are still unclear. In the central nervous system, the main histamine producer is within tuberomammillary nucleus which projects not only to vestibular nuclei but also thalamus,

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cortical areas, and others.[13] Recent studies suggested the role of histamine in modulation of vestibular nuclei neurotransmission, central synaptic plasticity, cognitive functions, and stress response.[13]

Anti-histaminergic compounds may probably facilitate vestibular compensation by assisting in the reduction of sensitivity to abnormal peripheral afferent.[14] For example, decreased gain of the horizontal which was reported after systemic treatment with histamine 3 (H3) reverse agonist thioperamide, as well as betahistine, another H3 receptor antagonist. If that is the case, even though this anti-histaminergic could decrease symptoms of BPPV, we suspected that such medication might impede a full recovery of vestibular function through VREs.

At Trang Hospital patients with BPPV were treated by otolaryngologist who prescribed medication and referred them for VR therapy. A BPPV rehabilitation clinic was established in 2007. Since then, there has been an average of 140 BPPV patients received VR therapy per year. Before therapy, each patient was assessed with the global BPPV symptoms (vertigo, dizziness and balance problem) severity assessment using a VAS (visual analog scale) diagram and a set of BPPV provocation test and vestibulo-ocular function tests as follows: the Dix-Hallpike test (DHT), the roll test (RT), the head thrust test (HTT) and the gaze evoked nystagmus test (GENT) (Appendix 1). All were carried out and recorded by physical therapist, the first investigator. The first assessment took place immediately befoe the beginning of the first therapy session. The second and the third assessments took place just before each weekly therapy session. The last assessment took place one week after the third therapy session. All assessments were carried out by the first investigator. According to our VR therapy (Appendix 2), the patients underwent one or another CRP technique, depending on an identified location of otolith in the semicircular canal. Then, they were guided through a series of VREs[15] which consisted of vestibulo-|ocular reflex (VOR) training with fixed target, VOR training with moving target and a side lying exercise (Brandt and Daroff exercise). All exercises were demonstrated by physical therapist, and the patients were informed to complete 4 sets of 3 repetitions of each exercise per day at home. All data of the assessments and the therapy were recorded in the case record forms.

In our previous retrospective pilot study, we have found that patients who took antihistamine medication showed less improvement of BPPV symptoms as measured with VAS. Betahistine has been the most commonly antihistamine prescribed for the treatment of BPPV symptoms at Trang Hospital.[16] To our best knowledge, there had never been a study focusing on the effect of betahistine on the recovery of these VOR related oculo-motor functions in those with BPPV. Therefore, the objective of this study was to investigate whether betahistine had a negative effect on recovery of brainstem VOR integration after completing three sessions of VR therapy and a daily home-based VREs for three weeks.

Methods

After obtaining the approval from the Trang Hospital Ethical Review Board (certification letter number 030/10-2562) the research was conducted as per the following details.

Participants

Data from medical records and case record forms (CRFs) of all patients who were referred to the BPPV clinic, the Division of Physical Therapy, for vestibular rehabilitation therapy, during October 2017 until August 2018 were retrospectively reviewed and analyzed.

Based on our pilot study which showed standard deviation of 7.99 and defining mean difference of 6.4, a sample size was calculated with software PS sample size: online available: www.Power-Analysis.com. As the result, 38 patients from each group (a group of taking betahistine and a group of not taking any antihistamine) in order to achieve statistical power of 0.8 and statistical significance at $p < 0.01$

Study protocol

From a total 525 medical records reviewed, 360 patients were excluded: 56 cases took other antihistamine medicine other than betahistine and 304 had incomplete data making them useless for analysis.

The BPPV assessment and the VR therapy case record forms (CRFs) of the recruited patients were selected and divided into two groups, those taking betahistine and those not taking any antihistamine. Then, the relevant data of the BPPV assessments/tests (Annex 1) before, during and after the therapy were retrospectively reviewed and extracted for analysis.

Statistical analysis

Demographic data were analyzed with descriptive statistic. Because the distribution of scores was not normally distributed, a non-parametric statistic test was used. Changes of BPPV tests scores across the course of VR therapy for each group were calculated with Friedman test. The difference of each assessment score between the betahistine and the no antihistamine groups were analyzed using Mann-Whitney test. Statistic calculations were done using MedCalc Statistical Software version 19.1 (MedCalc Software, Ostend, Belgium; https://www.medcalc.org; 2019)

Results

Of the 165 patients (37 males and 128 females) who completed the VR therapy and received all four BPPV assessments sessions necessary for analysis, there were 81 patients in the betahistine group and 84 in the no antihistamine group. Mean age was 58.26 (SD 13.15) years. Mean duration of BPPV symptoms was 34.71 (SD 34.05) days prior to the first visit. Table 1 shows comparisons of demographic data of the patients in the betahistine and the no antihistamine groups.

Table 1. Comparisons of demographic data of the patients in the betahistine and the no antihistamine groups

	Betahistine (n=81)	No antihistamine (n=84)
Gender[1]		
- Male	20 (24.7)	17 (20.2)
- Female	61 (75.3)	67 (79.8)
Age[2]	55.58 (13.14)	60.85 (12.81)
Duration of sickness[2] (days)	30.44 (32.64)	38.86 (34.69)
Comorbidities		
- Dyslipidemia (DLP)	5	6
- Diabetes mellitus (DM)	0	6
- Hypertension (HT)	5	8
- HT and DLP	8	9
- HT, DM, and DLP	1	12
- Others	8	10
- No comorbidities	54	33

[1]Number (%), [2]mean (SD)

BPPV, benign paroxysmal positional vertigo

Table 2. Data of median (IQR) of the scores from the four assessments sessions

Test		Betahistine		No antihistamine		*p*-value[b]
		Median (IQR)	*p*-value[a]	Median (IQR)	*p*-value[a]	
BPPV symptoms severity VAS	Pre	9 (9.00 to 10.00)		9 (9.00 to 10.00)		0.371
	Post 1	4 (3.00 to 5.00)	< 0.001	5 (4.00 to 6.00)	< 0.001	0.001
	Post 2	4 (3.00 to 4.00)		2 (2.00 to 3.00)		< 0.001
	Post 3	2 (1.00 to 2.00)		0.00 (0.00 to 0.00)		< 0.001
Dix-Hallpike test (DHT)	Pre	1 (1.00 to 1.00)		0.238 (0.00 to 0.00)		0.004
	Post 1	1 (0.00 to 1.00)	< 0.001	0.107 (0.00 to 0.00)	< 0.001	0.002
	Post 2	0.222 (0.00 to 0.00)		0.035 (0.00 to 0.00)		0.087
	Post 3	0 (0.00 to 0.00)		0.035 (0.00 to 0.00)		0.087
Roll Test (RT)	Pre	0.074 (0.00 to 0.00)		0.238 (0.00 to 0.00)		0.004
	Post 1	0 (0.00 to 0.00)	< 0.001	0.107 (0.00 to 0.00)	< 0.001	0.002
	Post 2	0 (0.00 to 0.00)		0.035 (0.00 to 0.00)		0.087
	Post 3	0 (0.00 to 0.00)		0.035 (0.00 to 0.00)		0.087
Gaze evoked nystagmus Test (GENT)	Pre	2 (2.00 to 2.00)		2 (1.00 to 2.00)		0.018
	Post 1	2 (2.00 to 2.00)	< 0.001	0 (0.00 to 0.00)	< 0.001	< 0.001
	Post 2	2 (2.00 to 2.00)		0 (0.00 to 0.00)		< 0.001
	Post 3	2 (1.00 to 2.00)		0 (0.00 to 0.00)		< 0.001
Head thrust Test (HTT)	Pre	2 (2.00 to 2.00)		2 (1.00 to 2.00)		0.010
	Post 1	2 (2.00 to 2.00)	< 0.001	0 (0.00 to 0.00)	< 0.001	< 0.001
	Post 2	2 (2.00 to 2.00)		0 (0.00 to 0.00)		< 0.001
	Post 3	2 (1.00 to 2.00)		0 (0.00 to 0.00)		< 0.001

BPPV, benign paroxysmal positional vertigo; VAS, visual analog scale; NT, not testable

Pre, before the first therapy session; post 1, before the second session; post 2, before the third session; and post 3, one week after the third session

[a]Friedman test comparing repeated measurement of the same group over time

[b]Mann-Whitney test comparison between the two groups: the betahistine and the no antihistamine groups

Table 2 shows median (IQR) of the scores from the four assessments sessions (pre - before the first therapy session, post 1 – before the second session, post 2 – before the third session, and post 3 – one week after the third session). There were significant improvements of every assessment score ($p < 0.001$) in both groups. Before the first therapy session, the BPPV symptoms severity VAS score (SD) of the betahistine and the no antihistamine groups, were 9.12 (0.73) and 9.22 (0.70) (p=0.38); however, in the second assessment were 4.17 (0.86) and 5.15 (1.21) ($p < 0.001$); in the third assessment were 3.53 (0.63) and 2.57 (3.32) ($p < 0.001$), and in the last assessment were 1.84 (0.64) and 0.03 (0.18), respectively ($p < 0.001$).

Regarding the DHT, the RT, the GENT and HTT, in the baseline assessment mean scores in the betahistine group were not significantly different from the no antihistamine groups ($p > 0.01$). However, there were significant differences between groups in the GENT and the HTT scores in all subsequent assessments, favoring the no antihistamine group over the betahistine group ($p < 0.01$). However, in the first week assessment, the DHT and the RT scores were significant differences between the two groups, favoring the

no antihistamine group; but in the last two weeks of assessments the between group differences did not reach significant level (see Table 2).

Discussion

This study showed improvements in all assessments of BPPV symptoms after VR therapy, compatible with known facts that BPPV symptoms remit spontaneously with time, and that the CRP and the VREs shorten the recovery time. [10-12] However, the difference between the betahistine group and the no antihistamine group has never been mentioned previously. In the second assessment the VAS was significantly lower in the betahistine group but thereafter the no antihistamine group had instead lower VAS scores, and BPPV symptoms free was found only in the no antihistamine group at the end of the therapy. This suggests that perhaps antihistamine could initially help to alleviate the symptoms but possibly reduce the positive effects of vestibular rehabilitation in the longer run.

When looking at tests that challenge VOR and voluntary gaze control and stabilization such as the GENT and the HTT, the no antihistamine group seemed to have a faster recovery than the betahistine group. Therefore, it is suspected that antihistamine medication may reduce the adaptive response to benefit neurological adaptation. However, such medication may have no significant effect on the sensitivity of provocation test as the DHT and the RT scores in the last two assessment sessions showed no between group differences. This is not surprising because these two tests serve to provoke BPPV symptoms in cases with free moving otolith inside the semicircular canal. But over time the otoliths might have been resorbed and the vestibular organ and nuclei might have developed a lower sensitivity to the remaining bits already.

There were few limitations of this study. Firstly, it was a retrospective study of the VR therapy guideline for BPPV at Trang Hospital which was a part of routine-to-research to improve the management. Although the CRFs were set from the beginning, all the assessments and therapy were carried out by the first investigator only. Other limitations were that patients' compliance to medication (duration and dose of betahistine) and patients' adherence to the home-based VREs, were not controlled or recorded. To prove that betahistine really impedes the vestibulo-ocular functions, one should conduct an assessor-blinded, randomized controlled trial. Indeed, there has been a research proposal published about such a research being planned.[17] Besides, a longer-term follow-up should be carried out so that effects of medication on prevention of recurrence could be studied.

In conclusion, betahistine seems helpful for BPPV symptoms reduction in the first one or two weeks of vestibular rehabilitation therapy. One should consider discontinuation of the medication to promote more effective vestibular rehabilitation as it demonstrated significantly less global symptom reduction, and less recovery of vestibulo-ocular reflex functions, than those not receiving any antihistamine.

Disclosure

Watcharin Tayati and Assistant Prof. Parit Wongphaet declare no conflict of interest of any kind.

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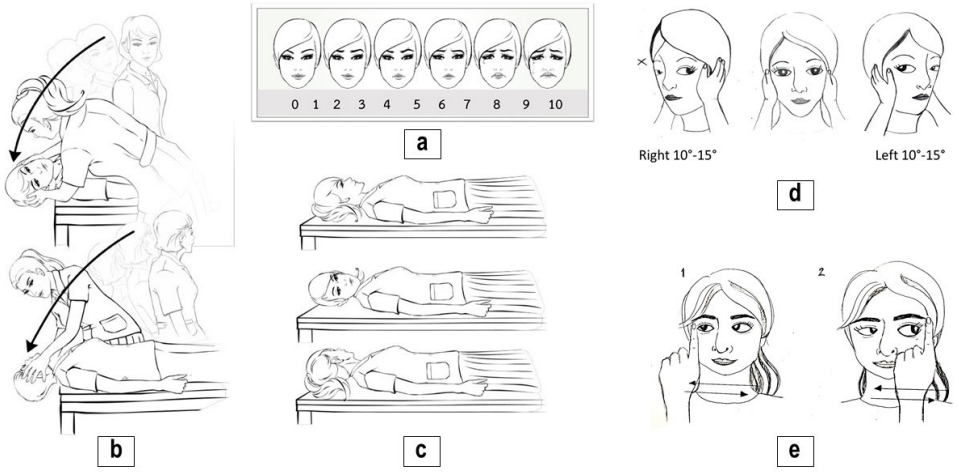
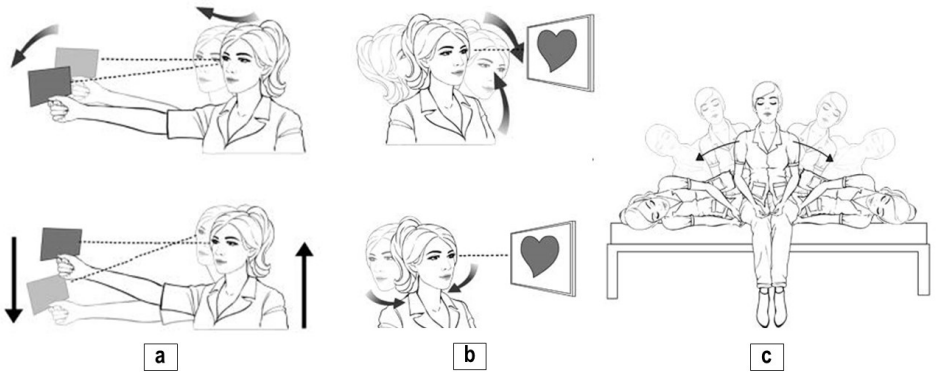
Appendix 1. BPPV assessments



a) Visual analog scale (VAS) diagram for global BPPV symptom severity, b) Dix-Hallpike test (DHT), c) Roll test (RT), d) Head thrust test (HTT), and e) Gaze evoked nystagmus test (GENT)

Visual Analog Scale: VAS score of zero indicates no problem at all and ten means the worst possible imaginable troublesome.
Dix Hall Pike test (DHP): A seated patient with neck turned 45 degree to one side is lowered quickly to a supine position with the neck extended 30 degrees below horizontal. The purpose of this test is to provoke symptom if there is otolith inside the anterior or posterior semicircular canal. (Score of 0,1, or 2 is given for each test when no, one sided, or bilateral nystagmus respectively.)
Roll test (RT): The neck of a supine lying patient is turned to one side, and then to the other side. The purpose of this test is to provoke symptoms if there is otolith inside the horizontal semicircular canal. (Score of 0,1, or 2 is given for each test when no, one sided, or bilateral nystagmus respectively.)
Head thrust test (HTT): A tester instructs a seated patient to fix his/her gaze on a target in front, then quickly rotate the head of the patient to one side about 10-15 degree, and then to another side. (Score of 0,1, or 2 is given for each test when no, one sided, or both eyes lose fixation to the target due to the passively induced quick short head turning respectively.)
Gaze evoked nystagmus test (GENT): A tester asks the seated patient to keep his/her head steady and fixes his/her gaze on a midline visual target which then was moved about 30 degree to one side, and then to another side.(Score of 0,1, or 2 is given for each test when no, one sided, or both eyes lost fixation to the target at any time.)

Appendix 2. Vestibular rehabilitation therapy program



a) Vestibulo-ocular reflex (VOR) training with a moving target, b) VOR training with a fixed target, and c) Side lying exercise (Brandt and Daroff Exercise)

Vestibulo-ocular reflex (VOR) training with a fixed target: starting from a straight sitting position with eyes fixing on a target in front. Then practice turning head back and forth horizontally or vertically while always keep looking at the target. The speed and amplitude of movement should be systematically and carefully increased without provoking a dizziness or vertigo.
Vestibulo-ocular reflex (VOR) training with a moving target: similar with the previous exercise, except that the target is being moved in the opposite direction with head turning. For example, when patient is turning the head from left to right the target is moved from right to left. This exercise aims to normalize influence of VOR on voluntary gaze control.
Side lying exercise (Brandt and Daroff Exercise): This exercise aims to desensitize the semicircular canal to the irritation of the otolith. Starting from a seated position facing the side of a bed then gently reposition into side lying position and remain in the position for 30 seconds.

Oculopharyngeal Muscular Dystrophy: a Case Report

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ABSTRACT

Objectives: To report rehabilitation management of oculopharyngeal muscular dystrophy (OPMD).
Study design: Case report.
Setting: Department of Rehabilitation Medicine, Maharat Nakhon Ratchasima Hospital.
Subject: A 72-year-old male who presented with progressive bilateral ptosis, hoarseness and swallowing difficulty, dysphagia.
Methods: Patient's medical records from 1st January 2018 to 31th December 2019 were reviewed and swallowing dysfunction was assessed by physical examination, a modified water swallow test and videofluoroscopy.
Results: Abnormal swallowing was found in the pharyngeal stage with decrease in laryngeal elevation and pharyngeal constriction. Penetration was seen after drinking a cup of water. OPMD was clinically diagnosed. Genetic confirmation of mutation in the PABPN1 gene was made. Nutritional assessment was performed. Swallowing rehabilitation program consisted of Shaker exercise and Mendelsohn maneuver to increase hyolaryngeal excursion, chin tuck for compensatory strategy and modified diet. The patient was discharged without complication. The swallowing program was continued and he was able to swallow safely.
Conclusion: OPMD is a rare disease causing dysphagia. Extensive family history-taking and physical examination leaded to diagnosis. Currently, there is no definite treatment for OPMD but proper swallowing rehabilitation management could improve swallowing dysfunction and improve patient's quality of life.

Keywords: oculopharyngeal muscular dystrophy, dysphagia, swallowing dysfunction, rehabilitation

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Introduction

Oculopharyngeal muscular dystrophy (OPMD) is one of nine types of muscular dystrophies.(1) Physical manifestation in patients begins with ptosis, which gets worse as time goes on. Later, the patients will have difficulty swallowing and at last will experience proximal limb weakness.(2) As the disease has its first manifestation in the voluntary muscles, OPMD is considered a rare disease. It is a genetic disorder, mostly from hereditary autosomal dominant which usually manifests in the fifth or sixth decade of life. However, some can also be an autosomal recessive disorder that happens rarely. It is difficult to diagnose as the manifestations happen at an advanced age and thus could be confused with aging conditions.(3) Therefore, the diagnosis requires genetic testing. Presently, the standard method for diagnosis is genetic study poly (A) binding nuclear protein 1 (PABPN1) gene in OPMD patients with autosomal dominant. It is found that there will be 12-17 repeats of expanded alleles. As for the autosomal recessive type, there are 11 repeats.(4)

The prevalence of autosomal dominant OPMD is high in Quebec province of Canada at the ratio of 1:1000 individuals. However, it was Isarael's Burkhara Jewish population that had the highest gene frequency at 1:600.(5) In Asian countries, there were some reports on the prevalence of OPMD, such as in China, Taiwan, Hong Kong, and Japan with reports on genetic confirmation in Southeast Asian countries such as Malaysia and Thailand.(6) Because it is a rare disease and not well-known outside Canada, specialists e.g. neurologists, otolaryngologists, have a potential to underdiagnose OPMD.(7) Though there has not been any exact report concerning the survival of untreated OPMD,(8) malnutrition and recurrent aspirations are important complications which can affect life expectancy of this group of patients if swallowing difficulty assessment is not included.(9) It is clear there will be great challenges in the diagnosis and treatment. Therefore, we would like to present a case with dysphagia in this rare genetic disease and results of swallowing assessment and rehabilitation program to improve swallowing function.

Case report

A 72-year-old Thai male with underlying type II diabetes and hypertension was presented to the hospital with hoarseness symptom for 6 years. Fiberoptic laryngoscope (FOL) examination was performed by an otolaryngologist, and presbylaryngis was diagnosed. He then lost to follow-up.

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One year later, he came with dyspepsia and bilateral ptosis. Gastritis was treated with proton pump inhibitors by an internist. His symptoms did not improve after being treated for two years. He was referred to gastro-enterologist for esophago-gastro-duodenoscopy (EGD) but findings were normal. Then the second FOL was done and showed only abnormal pooling of saliva at posterior cricoids, atrophic true vocal cord with good movement, and no mass in the oropharynx, pharynx and hypolarynx. The barium swallow showed normal visualized esophagus but large amount of barium suspension went into the trachea, thus the study was terminated. The patient needed oral feeding and his swallowing difficulty became worse. Finally, he was referred to a neurologist due to hoarseness, ptosis in both eyes and swallowing problems. He was asked to give a thorough record of his history as well as that of his extended family as shown in Figure 1.

His mother had an onset of symptoms, progressive dysphagia and bilateral ptosis, at the age of 57 years old, followed by an upper airway problem with stridor, and died of stroke at the age of 62 years old. His eldest brother had an onset of bilateral ptosis and ophthalmoparesis at the age of 66 years old and died of aspiration pneumonia at the age of 73. Another elder brother had uncertain onset of progressive bilateral ptosis and abnormal voice. The sixth brother had only bilateral ptosis and mild facial weakness at the age of 55 and died of metastatic brain cancer 7 years ago. The other two younger brothers presenting with bilateral ptosis, dysarthria, dysphonia and mild dysphagia at the age of 65 years old, were diagnosed with OPMD.

According to the above-mentioned family history, muscular dystrophy was suspected as the cause of his illness. The gene study was requested when he was referred to a rehabilitation physician (physiatrist) for assessment of swallowing difficulty. A DNA test reported heterozygous (GCG)9 or (GCN)13 expansion in the PABPN1 gene, resulting in an expansion of polyalanine tract from 10 to 13 residues in one allele, thus verifying the patient as having OPMD. Laboratory tests revealed normal serum albumin, creatinine and BUN levels, normal CBC counts and RBC indices, and HbA1C was 8.7%. His blood pressure was controlled. Muscle biopsy and serum creatine kinase level were not done.

Before admission for medical rehabilitation, his body weight decreased by 10% (from 55 kg to 48 kg) in the past year.

Dysphagia and ptosis progressed significantly. He reported difficulty swallowing of liquids primarily and then of solid foods but he denied ever experiencing choking events or lung infections. Physical examination showed good tongue strength and movement with no fasciculation or atrophy. Gag reflex was absent bilaterally. Soft palate elevation decreased on the left side and laryngeal elevation was more than one finger breadth when the patient performed a dry swallowing. Hoarseness was noticed but the patient could still communicate. The motor power was graded 5 in all extremities so he was independent in all daily activities and could walk without gait aid.

A modified barium swallow test showed decrease in laryngeal elevation and pharyngeal constriction and some residues after swallowing a tested food in an upright left lateral view (Figure 2), suggesting physiologic abnormalities in the pharyngeal stage, impaired relaxation of upper eso-phageal sphincter and inadequate airway protection. After doing a chin tuck, there was a decrease in residue after swallowing.

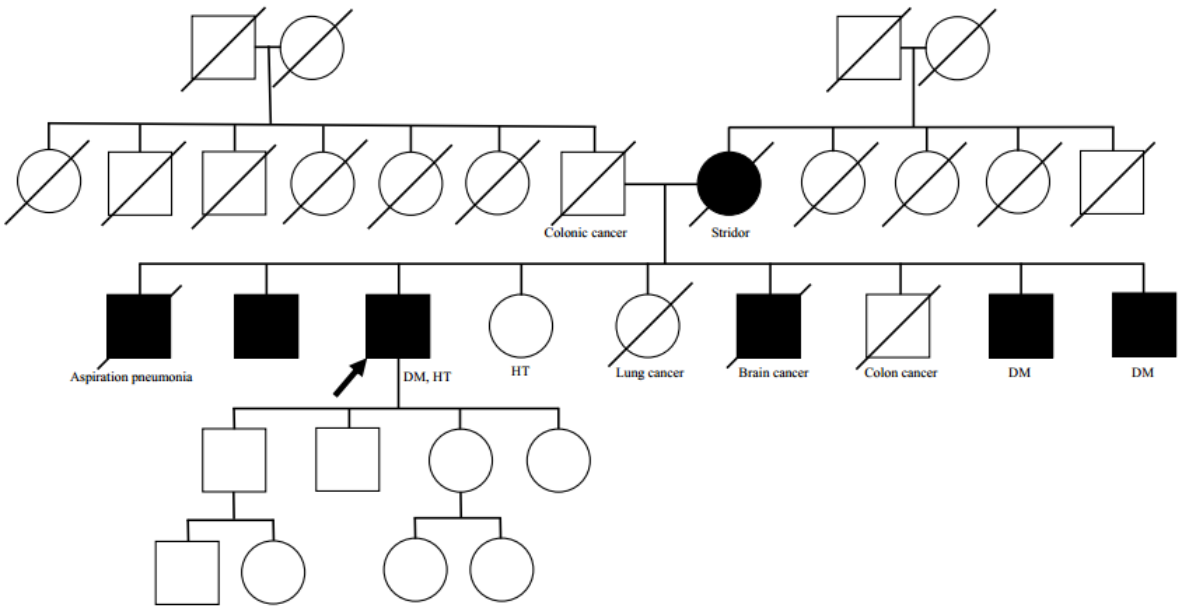

Figure 1. Pedigree of the patient's family: his mother (black circle) and six out of nine siblings including the patient (black square with black arrow) with diagnosis of oculopharyngeal muscular dystrophy (OPMD) indicating an autosomal-dominant transmission. The affection status of the maternal grandmother is unclear due to unrecognized history from the patient. The affection status of two generations after the patient are negative because the onset of symptom has not shown.

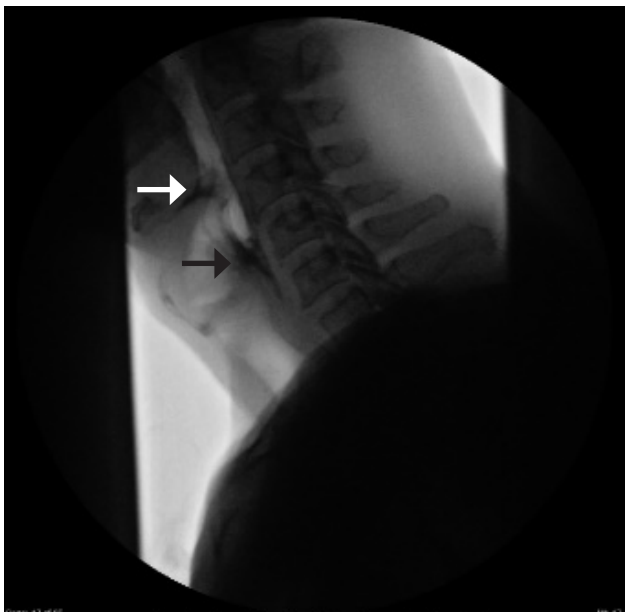

Figure 2. The modified barium swallow test shows residue in vallecular (white arrow) and pyriform sinus (black arrow).

Penetration was seen after drinking a cup of water and there was some residue after swallowing various food types such as liquid, paste, and solid. Oxygen saturation level did not change throughout the test.

During rehabilitation admission, the patient was trained to swallow using Mendelsohn maneuver and chin tuck position. In addition, Shaker exercise was advised to improve laryngeal elevation. Then he was discharged and asked to continue such program at home with a regular diet. After 2 months of home program, swallowing improved. Based on the Functional Oral Intake Scale (FOIS), swallowing was graded as level V, total oral diet with multiple consistencies, but required special preparations or compensations.

Discussion

Diagnosis of OPMD is difficult in Asian countries where the prevalence is low and even more difficult in cases of mild symptoms such as only ptosis or voice hoarseness. Multiple diagnoses and follow up visits were often made before getting a definite diagnosis of OPMD. In this case, he was firstly diagnosed by otolaryngologist because his main complaint was hoarseness. Only later when ptosis and swallowing difficulty increased, he was referred to a neurologist as a genetic neuromuscular disorder was suspected. Without thinking of this group of diseases, a misdiagnosis could lead to delayed treatments.

At present, treatments for OPMD are symptomatic, like those of degenerative neurological disease.[9] From previous researches,[1,3,9] swallowing difficulty started with solid food followed by liquid. This patient provided a different history as the problem started with liquid food and over 10% weight loss which led to a suspicion of pharyngeal muscle dysfunction. The modified barium swallow test showed impaired swallowing of both solid and liquid at the pharyngeal stage, similar to previous reports.[1,2,9,10,11] It was reported that 77.3% of patients with OPMD had food residues in vallecular and 90.1% had food residues in pyriform sinus. According to a review of OPMD, prevalence of penetration or aspiration was up to 33% depending on the degree of swallowing difficulty.[2] In this patient, penetration was found after drinking a large amount of water and had impaired laryngeal elevation in a forward position. Therefore, he was trained to swallow with the Mendelsohn maneuver to keep the upper esophageal sphincter open, perform Shaker exercise to strengthen suprahyoid muscle, and keep chin tuck while swallowing to narrow the respiratory tract. In addition, dietary modification was prescribed as reported[2,10] because patients with OPMD and swallowing problems at the pharyngeal stage usually have weakness of pharyngeal constrictors and hypertonia of the upper esophageal sphincter.[9,10] For those with clear symptoms of cricopharyngeal dysfunction, botulinum toxin injection of 0-100 units with electromyographic guidance and under general anesthesia should be considered before cricopharyngeal myotomy.[10,11] The latter might cause complications such as paralysis of vocal cord (unilateral), dysphonia, or progression of dysphagia, and a recurrence of swallowing problem after surgery was reported.[11] If there is dysphagia in the oral stage such as drooling, a protocol of improving oro-motor function should be added.[3]

Concerning swallowing difficulty in this case, if there had been an early assessment, an appropriate advice and proper treatments, it could decrease malnutrition and other potential and harmful complications. Therefore, an initial treatment focusing on swallowing rehabilitation program should include modified foods, compensation strategies and swallowing exercise program, tailored to individual swallowing problems; and placing importance on safety eating with nutritional assessment at the same time.

In conclusion, patients with oculopharyngeal muscular dystrophy may present with swallowing difficulty due to weakness of pharyngeal constrictors and/or hypertonia of the upper esophageal sphincter. Pedigree and family history-taking is important to establish a pattern of transmission and diagnosis. Since there is no definite treatment at present, swallowing rehabilitation program is still very much needed and prescribed by rehabilitation physicians (physiatrists).

Declaration conflict of interest

The authors has no conflict of interest to declare.

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