



Myung Sook Park, PhD, RN
Jin Yi Choi, PhD, RN
Young-Jin Song, MPH
Heejung Choi, PhD, RN
Eun-Jun Park, PhD, RN
Eun Sun Ji, PhD, RN



Systematic Review of Behavioral Therapy to Improve Swallowing Functions of Patients With Parkinson's Disease

ABSTRACT

Decreased swallowing function is a common and main cause of malnutrition and aspiration pneumonia in patients with Parkinson's disease. The aims of this systematic review were to summarize and qualitatively analyze the studies that have been published on behavioral therapies for improving swallowing functions in patients with Parkinson's disease. Studies published from January 2000 to December 2015 were identified via electronic database searches using Ovid-MEDLINE, Ovid-EMBASE, the Cochrane Library, and 8 Korean databases. Two reviewers independently evaluated the studies using inclusion criteria. Nine studies were included, of which 6 evaluated rehabilitation technique studies and 3 evaluated compensatory strategies. The 9 studies were evaluated qualitatively using a methodology checklist of the Scottish Intercollegiate Guideline Network, according to which all of the studies had acceptable quality. The available data on the effects of rehabilitation techniques and compensatory strategies remain insufficient. Further randomized controlled studies should be done to investigate the effect of behavioral therapy on improving swallowing functions in patients with Parkinson's disease.

Parkinson's disease (PD) is a neurodegenerative disease that occurs as a result of progressive loss of dopamine-secreting neurons in the substantia nigra of the brain. It is characterized by aggravating tremor at rest, stiffness, a posture that is mildly bent forward, slower conscious movements, and

a lack of facial expression that is known as masked face (Adams, Vitor, & Ropper, 1988; Chaudhuri, Healy, Schapira, & National Institute for Clinical Excellence, 2006). In the United States (U.S.), there were 630,000 patients with PD as of 2010, and it has been predicted that this number will double by 2050. It has also been reported that the treatment costs for patients with PD in 2010 were double those of patients in similar conditions without PD (Kowal, Dall, Chakrabarti, Storm, & Jain, 2013). In South Korea, the number of patients with PD rose rapidly from 30,000 in 2004 to about 74,000 in 2012, with the most marked increase occurring in those 40 years of age or older. Parkinson's disease results in considerable financial burdens, especially because it requires active treatment and nursing care. The annual medical treatment cost for PD exceeds 200 million Korean won (\$1.48 million U.S.) (National Health Insurance Service, 2013).

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About the authors: Myung Sook Park, PhD, RN, is Associate Professor, Department of Nursing, Konkuk University, Chungju, South Korea.

Jin Yi Choi, PhD, RN, is Assistant Professor, Department of Nursing, Konkuk University, Chungju, South Korea.

Young-Jin Song, MPH, is Occupational Therapist, Department of Occupational Therapy, Asan Medical Center, Seoul, South Korea.

Heejung Choi, PhD, RN, is Professor, Department of Nursing, Konkuk University, Chungju, South Korea.

Eun-Jun Park, PhD, RN, is Associate Professor, Department of Nursing, Konkuk University, Chungju, South Korea.

Eun Sun Ji, PhD, RN, is Associate Professor, Department of Nursing, Konkuk University, Chungju, South Korea.

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Correspondence to: Jin Yi Choi, PhD, RN, Department of Nursing, Konkuk University, 268 Chungwon-daero, Chungju, Chungcheongbuk-do, 27478, Republic of Korea (jinred75@kku.ac.kr).

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Background

Symptoms of PD are broadly classified into motor and nonmotor symptoms. Nonmotor symptoms such as reduced swallowing function, drooling, swallowing difficulty, and suffocation are common. Generally

occurring as a result of PD progression or the side effects of dopamine therapy, these symptoms are quite serious and may induce inappropriate nutritional status or aspiration pneumonia (Chaudhuri et al., 2006; Muzerengi, Contrafatto, & Chaudhuri, 2007). In PD, dysphagia can occur in all steps of the swallowing process (Chaudhuri, Tolosa, Schapira, & Poewe, 2014) and is associated with reduced tongue control, delayed swallowing reflex, laryngeal and pharyngeal muscle weakness, and postural instability (Plowman-Prine et al., 2009). The prevalence of dysphagia in patients with PD has been reported to be about 35%, but the actual prevalence of oropharyngeal dysphagia was about 82% as determined by objective swallowing tests (Kalf, de Swart, Bloem, & Munneke, 2012).

Although dysphagia symptoms may seem to be similar in patients with PD and those with stroke, PD and stroke are fundamentally different. Stroke is associated with diverse symptoms, depending on the location and severity of injury, and patients frequently experience natural recoveries from stroke. Many studies of patients with stroke have suggested that rehabilitative techniques produce neurophysiological changes that lead to the recovery of oral nutrition, and that various compensatory strategies reduce aspiration-related complications (Speyer, Baijens, Heijnen, & Zwijnenberg, 2010). However, further studies are required to determine whether the interventions that are used for dysphagia in stroke patients could produce the same effects for dysphagia in PD patients, in light of the progressive, variable, and neurodegenerative features of PD. Therefore, in order to provide effective nursing care for patients with PD, it is crucial to perform studies that investigate interventions for preventing swallowing dysfunction and enhancing swallowing functions in PD patients.

To date, most of the dysphagia guidelines that have been developed have focused on acute or chronic stroke patients (Wirth et al., 2013), and none have been directed toward PD patients. This is presumably attributable to the different presentations of stroke and PD: 30% to 50% of acute stroke patients experience dysphagia, and dysphagia therapy is considered an important aspect of rehabilitation because it has a direct relationship with mortality. On the contrary, more than half of all patients with PD show tremor as the initial symptom, and only about 10% show gastrointestinal symptoms (Mehanna, Moore, Hou, Sarwar, & Lai, 2014; Wirth et al., 2013). The prevalence of dysphagia in PD is even lower (most cases of dysphagia occur in later stages of PD) (Cersosimo et al., 2013). Nevertheless, treatments and nursing efforts are highly important for dysphagia, as it is a serious symptom that may induce malnutrition or aspiration pneumonia (Chaudhuri et al., 2006; Muzerengi et al., 2007).

There have been three systematic literature reviews pertaining to interventions for dysphagia in PD patients, one of which reviewed studies published prior to 2000 (Deane, Whurr, Clarke, Playford, & Ben-Shlomo, 2001), another of which examined the effects of deep brain stimulation after 2000 (Troche, Brandimore, Foote, & Okun, 2013), and the third of which systematically reviewed overall treatment of dysphagia (Baijens & Speyer, 2009; van Hooren, Baijens, Voskuilen, Oosterloo, & Kremer, 2014). However, these studies mostly focused on medical treatments, such as drugs or surgeries, and largely neglected behavioral therapies, which could be applied as nursing interventions.

Behavioral therapies can be broadly classified into two categories: rehabilitative techniques that produce changes by stimulating functions and compensatory strategies that produce temporary effects in consideration of safety. Rehabilitative techniques include methods that transform physiological temperament through exercise or sensory stimulation, such as exercise therapy, electric stimulation, temperature and sensory stimulation, and postural training via muscle tone control. On the contrary, compensatory strategies focus on safety by adjusting the viscosity of food or changing the pathway of food passage via temporary postural changes (Song et al., 2007). Swallowing-related maneuvers are considered compensatory strategies but are sometimes included in rehabilitative techniques, as they produce rehabilitation during long-term applications. In addition, various intervention methods are needed to enhance cognitive functions, create appropriate environments, maintain nutritional well-being, and prevent dehydration (Beom & Han, 2013). These interventions are usually performed by a professional occupational therapist at a rehabilitation center but can be taught and monitored by nurses every day, such as during meal times.

In this context, the present review was performed to examine the domestic and foreign studies that have focused on identifying the types and effects of behavioral therapies for dysphagia in patients with PD. The review specifically excluded drugs and surgical treatments and instead focused on interventions that could be performed and taught directly by a nurse in a clinical setting. We have undertaken this review in an attempt to contribute to evidence-based nursing by providing grounds for the nursing of dysphagia.

The objective of this systematic review was to identify the types of behavioral therapy for dysphagia in patients with PD and to examine their effectiveness. The specific objectives were as follows: first, we sought to identify the types of behavioral therapies that improve dysphagia in patients with PD; second, we sought to analyze the specific effects of these behavioral therapies for dysphagia in patients with PD.

Methods

Study Design

This study was a systematic review that analyzed the effects of behavioral therapies that were intended to improve swallowing functions in patients with PD.

Inclusion and Exclusion Criteria

Inclusion Criteria

The inclusion criteria were as follows: the subjects of this study were patients with PD who had dysphagia and drooling, which are typical nonmotor symptoms of PD. Behavioral therapy was selected as an intervention for these symptoms.

Exclusion Criteria

The following types of studies were excluded from this review: studies with overlapping content; studies written in languages other than Korean and English; studies conducted on nonhuman species; nonoriginal articles, such as studies published in review articles or conference proceedings; case studies with less than 10 cases or single-arm experimental studies with no control group; studies on patients without PD or patients without dysphagia, drooling, or sialorrhea (e.g., studies of patients with gastroesophageal reflux or stroke); studies that were not interventional studies on dysphagia in PD; and studies that examined deep brain stimulation and drug therapies as interventions for dysphagia in PD.

Data Search, Collection, and Analysis

Data Search

Before performing the main search of this review, we began by searching the Korea Education Research Information Service (KERIS) and PubMed databases for literature published from 2000 to 2015 under the search term “dysphagia,” in order to fulfill PICO (Population, Intervention, Comparison, and Outcome), which is the basic clinical question format for evidence-based nursing. A total of 3,217 articles were found. After excluding articles that were irrelevant to our survey of interventions for dysphagia, we reviewed 854 articles to select the subjects and interventions and to determine the search terms and search strategies.

For foreign literature searches, we used Ovid-MEDLINE, Ovid-EMBASE, and the Cochrane Library, which the Scottish Intercollegiate Guideline Network (SIGN) generally considers as required databases for literature searches (SIGN, 2015). The search period was set from January 1, 2000, to December 31, 2015, and we cross-searched “dysphagia” and the MeSH terms “deglutitional disorder” and “Parkinson disease,” which led to a total of 1,366 foreign articles. For

domestic literature searches, we used a total of eight online search databases: KoreaMed, National Library of Korea, National Assembly Library of Korea, Research Information Sharing Service by KERIS, Korean Medical Database, Korean Library Information System Network, National Digital Science Library, and Korean Studies Information. We used the search terms “dysphagia,” “swallowing difficulty,” and “swallowing problem,” and a total of 3,103 articles were found. However, because of the limited sensitivity and specificity of search features in the Korean databases, we did not cross-search PD; instead, we narrowed down our searches manually after performing a broad search using the aforementioned search terms.

Data Collection and Selection

The entire data collection and selection process was performed by two teams of two researchers (four researchers in total), each of whom had a minimum of 3 years of clinical experience and a PhD degree in nursing. All articles were reviewed independently, and when any disagreement occurred, the article under question was reviewed in depth with a third researcher.

A total of 4,469 articles (3,103 Korean articles and 1,366 foreign articles) were found via the search strategy. As per the primary screening, 2,128 articles were excluded because of overlapping content (1,894 Korean articles and 234 foreign articles). In addition, the following articles were also excluded: 99 articles written in languages other than Korean and English; 17 articles that examined nonhuman subjects; 846 articles published as review articles or conference proceedings; 484 articles that included less than 10 subjects or had a single-arm study design without a control group; 242 articles with subjects who did not have PD or dysphagia, drooling, or sialorrhea; 627 articles that were not interventional studies on dysphagia in PD; and 17 articles that examined deep brain stimulation and drugs as interventions for dysphagia in PD. As a result, a total of nine articles were selected for the final analysis (Figure 1).

Quality Assessment of Selected Literature

To determine the study design in an unambiguous manner, we began by performing a quality assessment using a SIGN algorithm (Shea et al., 2007). Subsequently, we assessed the quality of the selected articles (excluding single-group and single-group pre/post-test studies) based on the “methodology checklist” suggested by SIGN (Shea et al., 2007). The quality assessment was performed by two teams of two researchers (a total of four researchers) who assessed the studies independently and reached an agreement on the final determination. Articles that were determined

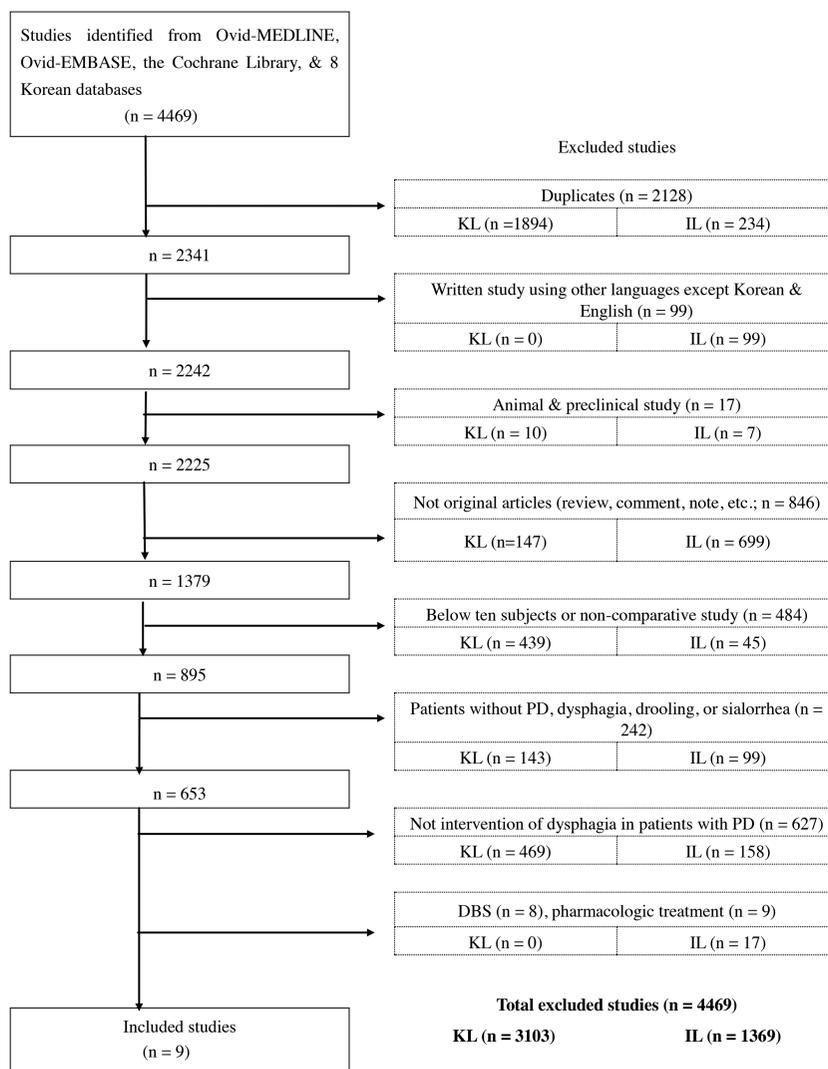


FIGURE 1. Flow diagram of article selection. DBS = deep brain stimulation; IL = international literature; KL = Korean literature; PD = Parkinson's disease.

to be above the acceptable level were selected for the final analysis (Figure 1).

Results

General Features of Selected Literature

A total of nine studies were selected for the final analysis, all of which were randomized controlled trials. However, there were problems in the randomizations of 7 (>70%) articles (Table 1). All nine articles were assessed as being acceptable (1+) according to the SIGN quality assessment. Three studies were published during 2000–2009 and six were published during 2010–2015; the number of interventional studies on dysphagia in PD has increased since 2010. Regarding the locations of the studies, three of the articles were published in the U.S., three were published in the Netherlands, and one was published in each of the

following nations: Israel, Japan, and Korea (Table 1). Six studies provided information regarding the severity of the subjects' PD on a Hoehn and Yahr (1967) scale (Baijens et al., 2012; Baijens et al., 2013; Heijnen, Speyer, Baijens, & Bogaardt, 2012; Manor, Mootanah, Freud, Giladi, & Cohen, 2013; Nagaya, Kachi, & Yamada, 2000; Troche et al., 2010). Most of the studies examined patients with Stage 4 PD or lower (Baijens et al., 2012; Heijnen, Speyer, Baijens, & Bogaardt, 2012; Nagaya, Kachi, & Yamada, 2000; Troche et al., 2010), but one study included patients in Stage 5, which represents severe PD in which patients require assistance for walking (Manor, Mootanah, Freud, Giladi, & Cohen, 2013). The remaining three studies did not indicate the severities of the subjects' PD (Kim, Lee, & Ha, 2012; Logemann et al., 2008; Robbins et al., 2008). Most studies had less than 100 subjects (Baijens et al., 2012; Baijens et al., 2013;

TABLE 1. Quality Assessment of the Selected Articles, Following the Scottish Intercollegiate Guideline Network Methodology Checklist

Type of Intervention	Author (Year)	Location	Study Design	Level of Evidence ^a	Appropriateness of Focused Question	Random Sequence Generation (Selection Bias)	Allocation Concealment (Selection Bias)	Blinding of Subjects (Performance Bias)	Blinding of Personnel (Performance Bias)	Blinding of Outcome Assessors to Therapy (Detection Bias)	Intra-Rater and/ or Inter-Rater Reliability Outcomes	Dropout Rate (%)	Intention to Treat Analysis
Swallowing exercise	Nagaya et al. (2000)	Japan	RCT	1+	Yes	Can't say	No	No	No	No	Yes	No comment	Can't say
	Manor et al. (2013)	Israel	RCT	1+	Yes	Can't say	Can't say	Can't say	Can't say	Can't say	Yes	No comment	Can't say
	Troche et al. (2010)	United States	RCT	1+	Yes	Can't say	Can't say	Yes	Yes	Yes	Yes	10	Yes
Electrical stimulation	Baijens et al. (2012)	Netherlands	RCT	1+	Yes	Can't say	No	No	Can't say	Yes	Yes	0	Does not apply
	Baijens et al. (2013)	Netherlands	RCT	1+	Yes	Can't say	Can't say	No	No	Yes	Yes	17.4	No
	Heijnen et al. (2012)	Netherlands	RCT	1+	Yes	Can't say	No	Can't say	Can't say	Can't say	Yes	19.3	No
Diet modification and postural adjustment	Kim et al. (2012)	South Korea	RCT	1+	Yes	Can't say	Can't say	Can't say	Can't say	Can't say	Yes	No comment	Can't say
	Logemann et al. (2008)	U.S.A.	RCT	1+	Yes	Yes	Yes	Can't say	Can't say	Can't say	Yes	4.2	No
	Robbins et al. (2008)	U.S.A.	RCT	1+	Yes	Yes	Can't say	No	No	No	Yes	2.1	Yes

Note. RCT = randomized controlled trial.

^aAll nine articles were assessed as being acceptable (1+) according to the SIGN quality assessment.

Heijnen, Speyer, Baijens, & Bogaardt, 2012; Kim, Lee, & Ha, 2012; Manor, Mootanah, Freud, Giladi, & Cohen, 2013; Nagaya, Kachi, & Yamada, 2000; Troche et al., 2010), but two studies examined more than 500 subjects (Logemann et al., 2008; Robbins et al., 2008). The ages of the subjects ranged from the fourth to the ninth decades of life.

Types and Effectiveness of Dysphagia Interventions for PD

In terms of types of behavioral interventions, the majority of the studies (six studies) investigated rehabilitative techniques, including three studies that examined swallowing exercise therapy (Manor, Mootanah, Freud, Giladi, & Cohen, 2013; Nagaya, Kachi, & Yamada, 2000; Troche et al., 2010) and three studies that examined electric stimulation therapy (Baijens et al., 2012; Baijens et al., 2013; Heijnen, Speyer, Baijens, & Bogaardt, 2012). The remaining three studies examined dietary and postural changes, which are considered compensatory strategies (Kim, Lee, & Ha, 2012; Logemann et al., 2008; Robbins et al., 2008) (Table 2). Because repeated applications of swallowing-related maneuvers induce increased intrapharyngeal pressure and changes in muscular contraction, they tend to be classified as swallowing exercises. Hence, we classified studies that applied swallowing-related maneuvers in addition to exercise therapy as swallowing exercise interventions (Nagaya, Kachi, & Yamada, 2000). The outcome variables were broadly divided into (1) swallowing function, (2) swallowing-related complications, and (3) quality of life and/or treatment quality assessments. First, swallowing function assessments were performed with endoscopic examinations, electromyography (EMG), and video fluoroscopy. Second, swallowing function-related complications were determined on the basis of the prevalence of pneumonia and aspiration, the penetration-aspiration scale for evaluating safety during swallowing, the functional oral intake scale, and body weight changes. Third, quality of life and/or treatment quality were assessed on the basis of the pleasure of eating, swallowing quality of life, and quality of life and treatment satisfaction using the MD Anderson Dysphagia Inventory.

Effectiveness of Swallowing Exercise Therapy in PD

There were three studies that examined rehabilitative techniques (specifically exercise therapies) for the recovery of swallowing function in patients with PD. Manor, Mootanah, Freud, Giladi, and Cohen (2013) conducted a study of 42 PD patients with dysphagia (H-Y scale score of 0–5), in which the control group ($n = 21$) underwent a swallowing exercise and compensatory strategy, whereas the experimental group ($n = 21$)

underwent video-assisted swallowing therapy for five 30-minute sessions over 2 weeks, in addition to the swallowing exercise and compensatory strategy. The results showed that both the control and experimental groups had significantly less pharyngeal residue as determined by a fiber optic endoscopic evaluation of swallowing ($p < .05$). There were no significant differences between the two groups ($p = .32$). However, the swallowing disturbances questionnaire scores, which probe the patients' perceived oropharyngeal swallowing problems, were significantly lower in the experimental group immediately after the intervention and 4 weeks after the conclusion of the intervention, as compared with the corresponding scores in the control group ($p < .05$). This finding indicates that adding video-assisted swallowing therapy to the traditional exercise and compensatory interventions reduced the patients' subjective swallowing disturbances. In addition, although there were no significant differences between the two groups in terms of Swallowing Quality of Life scores, the experimental group had significantly better mental pressure, appetite, social functions, mental health, and symptom expression than the control group. Swallowing quality of life significantly increased in both groups after the intervention.

Nagaya, Kachi, and Yamada (2000) studied 10 patients with PD and dysphagia (H-Y scale score of 3–4) who were 47–93 years of age, as well as 12 healthy adults in the same age range. Both groups were instructed to perform one set of an exercise program that applied four methods: exercise for adduction of the vocal cord; tongue range of motion (ROM) and resistance exercise; Mendelsohn maneuver; and neck, trunk, and shoulder ROM exercise. Swallowing-related parameters were assessed before and after the intervention using a video fluorographic swallowing study and EMG for both groups. During the test, the experimental group took levodopa, a dopamine drug, but did not take other drugs that affect the central nervous system. The results of the study showed that the experimental group (patients with PD) had significantly delayed pre-motor times, which represent the time that it takes to initiate the swallowing reflex, on the video fluorograph prior to the experimental intervention ($p = .001$), but that this delay decreased significantly after the exercise intervention ($p = .005$). The electromyographic burst, which measures the duration of the swallowing reflex of the muscle, did not vary significantly in the experimental group before and after intervention, indicating that the combined exercise program did not influence swallowing reflex.

In light of the fact that breathing and swallowing share various structures, such as the airway and relevant muscles, another study experimented with strengthening expiratory muscles, which can influence

TABLE 2. Summary of the Selected Articles on Behavioral Therapies for Dysphagia in Patients With Parkinson's Disease

Type of Intervention	First Author	Dopamine Medication	Subjects	Intervention/Exposure	Results
Swallowing exercise	Nagaya et al.	Yes	N = 22, mean age: 72 years H-Y scale score: 3–4 EG (n = 10) diagnosis: PD CG (n = 12) age-matched healthy volunteers	Swallowing training: Duration: 5 exercises. Only 1 session for both Content: ROM and resistance exercise of tongue; exercises for the adduction of vocal cords; Mendelsohn maneuver; ROM in the neck, trunk, and shoulder joints Measurement: BL, immediately post	Premotor time (EMG) at submental muscle by VFSS: EG (p = .005) Duration of EMG burst: n.s.
	Manor et al.	Unknown	N = 42, PD Group 1 (n = 21) Mean age: 67.6 years Group 2 (n = 21) Mean age: 69.9 years H-Y scale score: 0–5	Group 1: VAST Group 2: Swallowing exercises and compensatory therapy. No visual cues Duration: 5 times, 30 min, for 2 week Measurement: BL, immediately post, 4 weeks, 6 months	Swallowing function (by FEES): n.s. Swallowing disturbances: ↓ between the group at immediately post–4 weeks (p < .005), BL–immediately post (p < .001) and immediately post–4 weeks (p < .005) in G1 SWAL-QOL: n.s.
	Troche et al.	Yes	N = 60, Idiopathic PD H-Y scale score: 2–4 Range of ages: 55–85 years EMST, n = 30/CG, n = 30	EMST Duration: A weekly visit and home exercise for 4 weeks (5 days per week, 20 min per day) Measurement: BL, posttreatment	Swallow safety (PA scale): ↑ (p = .021) Swallow timing (VFS) of ten 5-mL thin swallow trials: n.s. EMST vs. ↓ CG (p = .007) Hyoid displacement at several swallow-specific events (VFS): ↑ all events in EMST (p < .01 in 3/7 events), ↓ CG (p < .05 in 4/7 events) SWAL-QOL: ↑ overall (p = .007)
Electrical stimulation	Baijens et al.	Yes	EG (n = 10) Idiopathic PD (5–13 years) Mean age: 66 years H-Y scale: 1–3 Dysphagic complaints: Mild to severe: MMSE ≥ 23 CG (n = 10) age- and gender-matched healthy control Median age: 65 years	Intervention Three different electrode positions (suprahyoid, infrahyoid, and bilateral combination) Random order per single-session protocol of SES during a standardized VFS swallowing protocol of 12 trials with thin liquid Measurement: SES during VFS	Electrode position: Significant effects Laryngeal vestibule duration(s): Infrahyoid < bilateral combination Duration of horizontal hyoid motion: infrahyoid > suprahyoid Effect of electrical current status (on/off): n.s. Significant group differences Velopharyngeal junction closure(s): EG > CG Laryngeal vestibule closure(s): EG > CG Laryngeal vestibule duration(s): EG < CG Upper esophageal sphincter closure (s): EG > CG Piecemeal deglutition (score of 0–4): EG > CG

(continues)

TABLE 2. Summary of the Selected Articles on Behavioral Therapies for Dysphagia in Patients With Parkinson's Disease (Continued)

Type of Intervention	First Author	Dopamine Medication	Subjects	Intervention/Exposure	Results
	Baijens et al.	Yes	<p>N = 90</p> <p>Idiopathic PD</p> <p>Dysphagic complaints: mild to severe</p> <p>Duration of PD: ≥5 years</p> <p>H-Y scale score of 1–5</p> <p>MMSE ≥23</p> <p>Mean age: 68 years</p> <p>Group 1 (n = 30)</p> <p>Group 2 (n = 30)</p> <p>Group 3 (n = 30)</p>	<p>Group 1: Traditional logopedic dysphagic treatment</p> <p>Group 2: Traditional logopedic dysphagic treatment + SES of the submental region (motor-level stimulation)</p> <p>Group 3: Traditional logopedic dysphagic treatment + SES of the submental region (sensory-level stimulation)</p> <p>Duration of intervention: 13–15 sessions of half an hour each, on five consecutive days per week within a period of 3–5 weeks</p> <p>Measurement: Immediately before and after therapy</p>	<p>Swallowing (FEES, VFS)</p> <p>Group differences (No SES effect): n.s.</p> <p>After therapy (before-after): ↑</p> <p>Piecemeal deglutition (OR = 0.73, CI = 0.60–0.89)/preswallow posterior spill for liquid bolus (OR = 0.49, CI = 0.27–0.89)/delayed initiation pharyngeal reflex (OR = 0.78, CI = 0.63–0.96)</p> <p>Bolus consistency effects</p> <p>Less piecemeal deglutition for thin liquid compared with crackers (OR = 7.043, CI = 4.54–10.90)/Less postswallow vallecular pooling for thin liquid compared with crackers (OR = 11.30, CI = 6.47–19.71)/Less preswallow posterior spill for thick liquid compared with thin liquid (OR = 0.30, CI = 0.10–0.89)/Less piecemeal deglutition for thin liquid compared with crackers (OR = 2.36, CI = 1.56–3.56)/Less delayed initiation pharyngeal reflex for crackers compared with thin liquid (OR = 0.35, CI = 0.22–0.54)/Less postswallow vallecular pooling for thin liquid compared with crackers (OR = 2.67, CI = 1.65–4.33)</p>
	Heijnen et al.	Yes	<p>N = 88</p> <p>Idiopathic PD</p> <p>Dysphagic complaints: Mild to severe</p> <p>H-Y scale: 1–4</p> <p>MMSE: 23–30</p> <p>Mean age: 68 years</p> <p>Group TT (n = 28)</p> <p>Group NMES-M (n = 27)</p> <p>Group NMES-S (n = 30)</p>	<p>TT: Traditional logopedic dysphagia treatment</p> <p>NMES-M: Traditional logopedic dysphagia treatment + NMES of the suprahypoid musculature on a motor level</p> <p>NMES-S: Traditional logopedic dysphagia treatment + NMES on a sensory level</p> <p>Duration of intervention: 13–15 sessions of half an hour each, on five consecutive days per week (median = 23 days)</p> <p>Measurement: BL, post, 3-month follow-up</p>	<p>DSS</p> <p>↓: TT ($p < .001$), NMES-M ($p < .001$), NMES-S ($p < .005$)</p> <p>Correlation between dietary intake and DSS: n.s.</p> <p>QOL</p> <p>n.s.: Group differences n.s.;</p> <p>↑ SWAL-QOL at post and follow-up</p> <p>MDADI: ↑ among total group and NMES-M ($p < .001$, $p = .007$)</p> <p>Correlations between SWAL-QOL, MDADI: n.s.</p>

(continues)

TABLE 2. Summary of the Selected Articles on Behavioral Therapies for Dysphagia in Patients With Parkinson's Disease
(Continued)

Type of Intervention	First Author	Dopamine Medication	Subjects	Intervention/Exposure	Results
Diet modification and postural adjustment	Kim et al.	Unknown	N = 50 Dementia, stroke, PD, others Mean age: 82.8 years EG: n = 25/CG: n = 25	EG: Posture, diet, & environment modification (4 wk.) CG: No program Measurement: BL, 4 weeks	Weight change ($p < .001$): EG ↑, CG Total swallowing time by EMG: EG ↓ ($p < .001$), CG n.s.
	Logemann et al.	Unknown	N = 711 PD without dementia, PD with dementia, dementia only Mean age: 50–95 years	Interventions: Chin-down posture while consuming thin liquids (CD), no postural adjustment during swallows of nectar-thickened liquid (NT), no postural adjustment during swallows of honey-thickened liquids (HT) Measurement: Simultaneous measurement	Aspiration frequency (VFS) PD only: CD > HT ($p < .001$); NT > HT ($p < .001$) PD with dementia: CD > HT ($p < .001$); NT > HT ($p < .001$) No correlation with PD severity (H-Y scale) Preference of intervention in PD without dementia: CD > HT ($p < .05$); NT > HT ($p < .01$)
	Robbins et al.	Unknown	N = 515 PD, dementia Mean age: 50–95 years Chin-down posture (n = 259) Nectar-thick liquid (n = 133) Honey-thick liquid (n = 123)	Chin-down posture drinking thin liquids Thick liquids in a head-neutral position Nectar-thick or honey-thick liquids Measurement: Until 3 months or death	Three-month cumulative incidence of pneumonia: 11% Incidence in chin-down posture group vs. thick liquids group: n.s. Incidence in nectar-thick liquids group vs. honey-thick liquids group: n.s.

Note. BL = baseline; CG = control group; CI = confidence interval; EG = experimental group; EMG = electromyography; EMST = expiratory muscle strength training; FEES = fiber optic endoscopic evaluation of swallowing; H-Y scale = Hoehn & Yahr scale; MDADI = M. D. Anderson Dysphagia Inventory; MMSE = Mini-Mental State Examination; NMES-M = neuromuscular electrical stimulation-motor level; NMES-S = neuromuscular electrical stimulation-sensor level; n.s. = not significant; OR = odds ratio; PA = penetration-aspiration; PD = Parkinson's disease; QOL = quality of life; ROM = range of motion; SES = surface electrical stimulation; SWA-CARE = swallowing quality of care; SWAL-QOL = swallowing quality of life; VAST = video-assisted swallowing therapy; VFS = video fluoroscopy.

swallowing functions. Troche et al. (2010) conducted expiratory muscle strength training using equipment on PD patients 55–85 years of age (H-Y scale of 2–4). Of a total of 60 patients, 30 underwent the training for five 20-minute sessions per week for 4 weeks, whereas the remaining 30 patients did not receive any type of intervention. In the experimental group, the penetration or aspiration scale values, which represent swallowing safety, were significantly higher after the intervention than before the intervention ($p = .021$). Although there were no evident changes in the video fluorograph results, the control group took a longer time to swallow food. In the experimental group, the degree of displacement of the hyoid bone (for safe swallowing) also increased significantly after the intervention. In contrast, this displacement decreased in the control group. Our quality assessment indicated that all three of the aforementioned articles were acceptable (1+).

Effectiveness of Electrical Stimulation Therapy for Swallowing in Patients With PD

Among the various rehabilitative techniques, a total of three studies examined electrical stimulation therapy, all of which used surface electrical stimulation. Baijens et al. (2012) conducted VitalStim therapy (frequency: 80 Hz, pulse width: 700 μ s; DJO Global, Vista, California) in 20 early- to mid-stage PD patients with dysphagia (H-Y scale score of 1–3) and 10 healthy controls of the same age group in random order. Electrodes were attached to the subjects' suprahyoid, infrahyoid, or both suprahyoid and infrahyoid bones, and swallowing function was measured using video fluorography. The results showed that velopharyngeal junction closure, laryngeal vestibule duration, upper esophageal sphincter closure, and piecemeal deglutition all increased significantly after the intervention in the experimental group, as compared with the control group. Furthermore, the effects of electrical stimulation were compared according to the placement of the electrodes, which showed that the laryngeal vestibule duration was significantly decreased for electrodes attached on both the suprahyoid and infrahyoid bones, as compared with electrodes attached on the infrahyoid bone alone ($p < .05$). In addition, the duration of horizontal hyoid motion was significantly increased for electrodes attached on the infrahyoid bone alone, as compared electrodes on the suprahyoid bone ($p < .05$).

Baijens et al. (2013) randomly divided 90 PD patients with dysphagia (H-Y scale score of 2–5) into three groups (traditional treatment, electrical stimulation of submandibular motor muscles, and electrical stimulation of submandibular sensory muscles) and applied corresponding interventions five times per week for 3 weeks (15 sessions). Swallowing endoscopic

testing and video fluorography showed no intergroup differences in swallowing function before and after the intervention, but intragroup comparisons showed enhanced swallowing functions after the intervention in all three groups.

Heijnen, Speyer, Baijens, and Bogaardt (2012) also conducted VitalStim Therapy (frequency: 80 Hz, pulse width: 700 μ s) for an average of 23 days in 88 PD patients with dysphagia (H-Y scale score of 1–4) who were randomly divided into three groups (traditional treatment, electrical stimulation of suprahyoid motor muscle, and electrical stimulation of suprahyoid sensory muscle). When the severities of dysphagia were compared before, immediately after, and 3 months after intervention, the three groups showed no intergroup differences. However, intragroup improvement was evident over time. Similarly, although intergroup differences were not observed for swallowing quality of life, improvement over time was seen within the groups. Our quality assessment of the aforementioned articles indicated that all three were acceptable (1+).

Effects of Dietary Changes and Postural Adjustments on Swallowing in Patients With PD

A total of three studies investigated dietary and postural changes as compensatory strategies. Logemann et al. (2008) studied 228 patients who had PD without dementia, 132 patients who had PD with dementia, and 315 patients who had dementia alone (total $n = 711$). The patients were divided into three groups (chin-down posture while consuming thin liquids, no postural adjustment while swallowing nectar-thickened liquid, and no postural adjustment while swallowing honey-thickened liquids) to measure the incidence of aspiration while eating food. The results showed that the use of a chin-down posture while consuming thin liquids was associated with a higher incidence of aspiration than the use of no postural adjustment while swallowing honey-thickened liquids. In addition, the use of no postural adjustment while swallowing nectar-thickened liquids was associated with a higher incidence of aspiration than the use of no postural adjustment while swallowing honey-thickened liquids.

Robbins et al. (2008) divided 515 patients who showed aspiration on thin liquids and had either PD or dementia into a chin-down group ($n = 259$), a nectar-thickened group ($n = 133$), and a honey-thickened group ($n = 123$). After monitoring their patients' diets for 3 months, Robbins et al. found no differences in the incidences of pneumonia among the groups. The studies by Logemann et al. (2008) and Robbins et al. (2008) were similar in that they did not measure the severity of PD, included patients with dementia, and used dietary

changes and postural adjustments as major interventions. However, the effects of the interventions varied according to the outcome variables. This is presumably explained by the idea that although compensatory interventions were found to be ineffective when pneumonia was the only outcome variable (Robbins et al., 2008), the thickness of food (rather than postural adjustment) was found to have a greater effect on the incidence of aspiration when the outcome variable was the incidence of aspiration during food intake.

In another interventional study, compensatory strategies were applied at a long-term care facility in Korea (Kim, Lee, & Ha, 2012). Fifty elderly patients with PD, dementia, and/or stroke were divided into experimental ($n = 25$) and control ($n = 25$) groups and taught a complex educational intervention that included an environmental intervention with postural adjustment for the prevention of aspiration. Teaching occurred for 10–13 hours for more than 4 weeks and was provided by their nursing care provider. As compared with pre-intervention values, body weight increased in the experimental group after 4 weeks but decreased in the control group over the same period. As measured by electromyogram, the total swallowing duration did not change in the control group but decreased in the experimental group after the intervention.

The studies by Logemann et al. (2008) and Robbins et al. (2008) reported that the following were effective in reducing swallowing time and increasing body weight: postural and dietary (thickness of food) adjustments, as well as an educational intervention encompassing a complex compensatory strategy that included environmental interventions such as cognitive stimulations and the use of a reduced stimulatory environment to help patients focus on eating. All three of the aforementioned articles on compensatory interventions were evaluated as having acceptable quality (1+).

Discussion

In this study, we have systematically reviewed studies published during 2000–2015 to identify the types and effectiveness of behavioral therapies for dysphagia in patients with PD. A total of nine articles were selected for review. The interventions from the reported studies were categories of rehabilitative therapies (such as swallowing exercise therapy and electrical stimulation therapy) or compensatory strategies (such as dietary and postural adjustments).

Of the six reviewed studies that focused on rehabilitative therapies, three examined swallowing exercise therapies. The types of the exercises varied, including (1) a complex program that consisted of exercise for adduction of the vocal cord; tongue ROM and resistance training; Mendelsohn Maneuver; and neck, trunk, and shoulder ROM (Nagaya, Kachi, & Yamada,

2000); (2) expiratory muscle strength training (Troche et al., 2010); and (3) video-assisted swallowing therapy (Manor, Mootanah, Freud, Giladi, & Cohen, 2013). Study subjects varied in terms of the severity of PD (H-Y scale score of 1–4), and the frequencies and durations of intervention administration varied as well.

Video-assisted swallowing therapy (Manor, Mootanah, Freud, Giladi, & Cohen, 2013) is a type of biofeedback therapy, which is a therapy that provides muscle re-education, relieves muscle tetany, and enhances voluntary control through visual and audio feedback, providing changes in neurophysiological functions in damaged motor patterns. Video-assisted swallowing therapy (Manor, Mootanah, Freud, Giladi, & Cohen, 2013) incorporated a biofeedback technique during exercise therapy by showing the subjects the passage of food through an endoscope, which reduced pharyngeal residue and improved some items of swallowing quality of life after the intervention. In general, multilateral studies are needed on the use of smart devices, such as videos and sensors, especially because smart devices have gained traction as a useful therapeutic technique that produces scientific and objective statistical results.

The variables assessed in studies of swallowing exercise interventions for PD have included premotor time of the submandibular muscle, as evaluated using video fluorography, EMG, and fiber optic endoscopic evaluation testing. Exercise interventions have been found to be effective for improving oral swallowing disturbance, improving objective and subjective swallowing functions, and preventing aspiration (Manor, Mootanah, Freud, Giladi, & Cohen, 2013; Nagaya, Kachi, & Yamada, 2000; Troche et al., 2010). However, one of the reviewed studies (Nagaya, Kachi, & Yamada, 2000) studied only 10 subjects and administered one session of the intervention, which hinders the generalization of their results. Furthermore, although Troche et al. (2010) reported the effects of an intervention after 4 weeks of treatment, further studies would be needed to observe the long-term effects of treatment, especially because PD progresses over a long period. In addition, the three studies did not control for the administration of levodopa, dopamine, or anticholinergic agents, presumably because dysphagia in PD can be related to both the progression of the disease and side effects of dopamine therapy. Furthermore, the three studies included heterogeneous severities of PD (Chaudhuri et al., 2006; Muzerengi et al., 2007).

Electrical stimulation therapy was investigated in the remaining three of the six reviewed studies on rehabilitative therapies (Baijens et al., 2012; Baijens et al., 2013; Heijnen, Speyer, Baijens, & Bogaardt, 2012). There are two types of electrical stimulation, one of which involves placing electrodes within the muscles and the other of which involves placing electrodes on

the surface of the skin (surface electrical stimulation). All three of the reviewed studies used the latter technique. The severities of PD were diverse (H-Y scale score of 1–5), and surface electrical stimulation was performed with a consistent intensity and frequency via VitalStim therapy (frequency: 80 Hz, pulse width: 700 μ s). The electrode placements and frequencies of stimulation varied across all three studies. Electrodes were placed on the suprahyoid bone, the infrahyoid bone, both the suprahyoid and infrahyoid bones, the suprahyoid motor muscle, the suprahyoid sensory muscle, the submandibular motor muscle, and the submandibular sensory muscle. The frequency of electrical stimulation ranged from one to 15 sessions. Evaluation variables included swallowing function testing via video fluorography and swallowing quality of life. There were no postintervention intergroup differences, but swallowing function and quality of life improved over time within the groups. Electrical stimulation is effective for subjects who have injured control of the central nervous system but whose nerves and muscles for swallowing are intact. Accordingly, it seems to have improved swallowing in patients with PD via contractions of the submandibular muscles, which lifted the pharynx because of displacement of the hyoid bone to the anterior and superior (Song et al., 2007).

Three studies examined the effects of diet modification and postural adjustment as compensatory strategies (Kim, Lee, & Ha, 2012; Logemann et al., 2008; Robbins et al., 2008). The diet modifications were the use of nectar-thickened or honey-thickened liquids, and posture was either adjusted to a chin-down position or was not adjusted at all. The durations of intervention varied from 4 weeks to 3 months. Evaluation variables included the incidence of pneumonia, incidence of aspiration, and degree of body weight change. From a nutritional perspective, body weight increased and swallowing time decreased, but the incidences of pneumonia and aspiration were not changed. Two studies applied the same compensatory strategy and found that the incidence of aspiration was lower in the nectar-thickened liquid group than in the chin-down group, and in the honey-thickened group than in the nectar-thickened group (Logemann et al., 2008; Robbins et al., 2008). This is presumed to indicate that compensatory techniques do not permanently improve the subjects' swallowing functions by inducing neurophysiological changes; rather, they bring about transient effects by changing the passageway or slowing down the food for swallowing (Beom & Han, 2013). Furthermore, changing the viscosity of the food was observed to be more effective than adjusting posture (chin-down), which is believed to be related to the fact that it is highly difficult for patients with PD to maintain their adjusted posture while eating. It is difficult

for patients to continuously maintain the corrected posture because of muscle imbalance, such as tremor and stiffness, as well as cognitive problems (Song et al., 2007). The study from Korea did not assess the progression or severities of PD and included multiple subjects with dementia or stroke, which makes it difficult to identify the effects of the intervention on PD, specifically (Kim, Lee, & Ha, 2012). Nevertheless, when postural adjustment and dietary modifications were both applied, continuous effects were observed even 4 weeks after the intervention (Kim, Lee, & Ha, 2012). This tends to indicate a synergistic effect between the two interventions, suggesting that further studies are warranted on complex compensatory strategies, rather than on single-approach strategies.

The present study classified behavioral therapies for dysphagia in PD into rehabilitative therapies (such as swallowing exercise therapy and electrical stimulation) and compensatory strategies (such as dietary modification and postural adjustment), which were similar to the behavioral therapies for dysphagia in stroke patients (Geeganage, Beavan, Ellender, & Bath, 2012). However, whereas studies on interventions for dysphagia in stroke patients reported various nutrition supply pathways, including percutaneous endoscopic gastrostomy and nasogastric tube, studies on interventions for dysphagia in PD have mostly focused on the oral ingestion of nutrition (Geeganage et al., 2012). This is because dysphagia in stroke patients is a temporary symptom and complication that occurs early in the course of the disease, and the aims of the intervention are to convert the nutrition pathway from parenteral to oral pathways and to reduce complications and mortality associated with malnutrition and aspiration (Speyer et al., 2010). On the contrary, dysphagia in PD is a symptom or side effect that presents in later stages of the disease, and interventions aim to reduce aspiration, improve nutrition, and improve quality of life (Gazewood, Richards, & Clebak, 2013). In resemblance to the findings reported in systematic reviews of the literature on dysphagia in stroke, we have found that the currently available evidence is not sufficient to support behavioral therapy for dysphagia in PD. Nevertheless, the incidence of PD is increasing, and interventions for dysphagia in PD have been observed to reduce aspiration pneumonia, improve nutritional status, and help improve quality of life. Therefore, we suggest that further studies should be conducted on behavioral interventions for dysphagia and PD, and that these studies should be limited to patients with PD.

Implications for Practice

This systematic review examined behavioral therapies for dysphagia in PD, focusing on interventions that

could be performed and taught directly by a nurse in a clinical setting. The main behavioral therapies that have been applied for dysphagia in PD include swallowing exercise therapies, electrical stimulation therapies, dietary modifications, and postural adjustments during swallowing. Overall, the currently available evidence is not sufficiently robust to support behavioral therapy for dysphagia in PD. Nevertheless, several of the reported interventions were observed to reduce aspiration pneumonia, improve nutritional status, and help improve quality of life.

Conclusion

Our review showed that the most popular behavioral therapies for dysphagia in patients with PD have been rehabilitative techniques, such as exercise therapy and electrical stimulation, and compensatory strategies, such as diet modification and postural adjustment. Sufficient evidence is currently lacking to support the effects of these therapies. Nonetheless, multiple studies with acceptable (1+) quality have been published on exercise therapy and electrical stimulation, and the effectiveness of each of these therapies has been partially verified. In addition, three published articles (including one Korean article) have examined compensatory strategies that could be applied by nurses in the clinical setting. However, because these studies did not limit their subjects to patients with PD and produced different results, additional randomized studies are warranted to evaluate compensatory strategies for patients with PD, specifically. ★

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