

Review Article





Dysphagia in Parkinson's Disease: Prevalence, Impact and Management Challenges

Abstract

The aim of this review is to present an update of the prevalence, impact and management challenges of dysphagia in Parkinson's disease. In the last 5years, the prevalence of dysphagia in Parkinson's disease (PD) has been reported with a wide range of values in the literature. The dysphagia prevalence ranged from 11% to 100% of the patients with PD. This variation, generally, depended on the disease stage and the procedure of swallowing assessment. In the management of dysphagia, compensatory techniques, rehabilitation exercises and deep-brain stimulation have been conducted. Despite of the lack of high scientific evidences of efficiency of the majority of these interventions, the positive aspects and benefits are described in the actual literature. Moreover, this review paper discuss about the importance of the dysphagia assessment and treatment in the best timing to prevent complications, as pulmonary and nutritional impairment, and to improve quality of life.

Keywords: parkinson's disease, swallowing, dysphagia, prevalence, treatment

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Abbreviations: PD, parkinson's disease; VFSS, video fluoroscopic study of swallowing; UKPDBB, united kingdom parkinson's disease brain bank; SDQ, swallowing disturbances questionnaire; UPDRS, unified pd rating scale; ASHA, american speech language hearing association; SLP, speech language pathologist; FEES, fiber optic endoscopic evaluation of swallowing; EMST, expiratory muscle strength training

Dysphagia in parkinson's disease

Neurological disorders may manifest a range of symptoms, as muscle weakness, hypotonia or hypertonia, hyperkinesia or hypokinesia, presence of involuntary movements, and lack of movement's coordination. The alteration of muscular condition and movement's coordination may affect the efficiency of the swallowing process, leading to dysphagia.

Oropharyngeal dysphagia refers to the alteration of the swallowing process from the intake of food in the mouth to the stomach. The complications from oropharyngeal dysphagia can be related to pulmonary and nutritional aspects. The most common complaints of dysphagia are: the difficult to chew, inability to transport the food to the pharynx, sensation of food stucked in the throat, and the presence of coughing and choking before, during or after swallowing. However, depending on the peripheral or central sensory impairment, the patient may not have complaints, which may lead to a silent aspirations and/or pharyngeal stasis. Because of this, it is extremely important to identify risks that may be caused by the neurological disease in the swallowing process and perform structural and functional assessments, even in the absence of complaints.

Dysphagia is a common symptom in patients with Parkinson's disease (PD) and, generally, affects the majority of the patients during the natural course of the disease. It can be due to the degeneration of brainstem, basal ganglia circuits, in addition to cortical areas responsible to modulate swallowing function. It is important to note that often the patient with PD will not identify the presence of penetration, and cough will be inefficient to allow airway protection. Because of that, it is highlighted the importance of conducting instrumental evaluations to determine the rehabilitation techniques,

consistency, and amount of food in order to establish a feeding without pulmonary complications and nutritional risks.³

Prevalence of dysphagia in parkinson's disease

In the last 5years, the prevalence of dysphagia in Parkinson's disease (PD) has been reported with a wide range, from 11% to 100%. 4-16 The procedures used to assess the presence of dysphagia and its severity it also differs in studies. All the data from the studies, here considered, reporting the prevalence of dysphagia in PD, can be found in the Table 1. The study of Aydogdu et al.,4 assessed the prevalence of dysphagia using the Video fluoroscopic Study of Swallowing (VFSS) and the "United Kingdom Parkinson's Disease Brain Bank" (UKPDBB) guidelines to diagnose Parkinson's disease. In that study, 23 patients were identified with PD and underwent VFSS evaluation, from the total sample, 16 patients were diagnosed with dysphagia. However, Han et al.,5 study, which observed the prevalence of dysphagia considering Hoehn and Yahr (H&Y) stages of PD, using a questionnaire to assess dysphagia, the "Swallowing Disturbances Questionnaire" (SDQ) in 127 patients diagnosed with PD, revealed in stage 1 the prevalence was of 14.3%; in the stage 2 was of 16.7%; in the stage 3 was of 33,3%; in the stage 4 was of 50.0%; and in the stage 5 the prevalence of dysphagia was in 100.0% of the patients.

Perz Llorett et al.,6 used the questionnaire "Unified Parkinson's Disease Rating Scale (UPDRS)" to verify the presence of dysphagia. They found that 18% of the sample of 419 PD patients had dysphagia. Different values of prevalence were found in the study of Ou et al.,7 which had 36.3% of the PD patients with dysphagia, of a total the 518 PD patients assessed by using a range of questionnaires, as the "Unified PD Rating Scale (UPDRS) part III". In the same way, Walker et al. 8 reported a prevalence of 32.0% of dysphagia in the sample of 72 PD patients. Two questions were used to determine dysphagia "Do you have difficulty swallowing food or liquid or tablets? and Do you cough after eating/drinking?" and the questionnaire UPDRS.

In the literature, it also can be found studies 9-16 reporting swallowing difficulties, complications, concerns, aspiration, penetration and other symptoms. In the research of Argolo et al., 69 patients with PD underwent VFSS, of those 18.84% had lingual

pumping, which is usually associated with dysphagia. Akbar et al., ¹⁰ assessed 5,665,710 medical records of patients diagnosed with PD, of which 3.8% had been hospitalized with aspiration pneumonia (AsPNA). In the study of Cereda et al., ¹¹ the medical records of 6,462 patients with PD were also assessed and a prevalence of 11.7% of swallowing disturbances was found.

Five other studies, ^{12–16} that used the VFSS to assess the swallowing in PD patient, were found in this literature review. Kim et al., ¹² evaluated 33 patients and observed that 78.8% had clinical sign of penetration or aspiration. Rajaei et al., ¹³ had a prevalence of 45.5% for penetration and as aspiration in the sample of 33 PD patients. The authors Silverman et al., ¹⁴ found in its 68 PD patients, a prevalence

of 52.9% of swallowing concerns. In the study by Monteiro et al., 15 the prevalence of swallowing complaints was of 40.0% in PD patients. Lastly, one other study by Michou et al., 16 also reported prevalence of swallowing disturbances in eleven (50.0%) of the PD patients evaluated. Thus, the prevalence of dysphagia in Parkinson disease still needs to be investigated. At this moment, it is clear that clinical conditions, age, disease stage and the procedure of swallowing assessment could interfere on the results. However, it is obvious that the patients will develop swallowing problems during the progressive course of the disease 17 and, as consequence of the absence of complaints in the presence of signs of dysphagia, clinical and instrumental assessments of swallowing are recommended.

Table I Prevalence of dysphagia in patients with Parkinson's disease, considering publications of the last 5 years in PubMed database

Author, Year	Total Sample (n)	Age (years)	Stages of PD (Hoehn & Yahr) (Mean±Standard Deviation)	Type of assessment	Prevalence of Dysphagia (%)
Akbar et al. [10]	5,665,710	77 ± 0.08	NA	Medical Records	3.8% had AsPNA
Argolo et al. [9]	69	63.36 ± 11.62	2.3 ± 0.90	VFSS	18,84% exhibited LP
Aydogdu et al. [4]	23	65.7	1.9	VFSS	69,56% had dysphagia*
Cereda et al. [11]	6462	53 - 68	I-SI/ 2-S2/ 3-S3/ 4-S4/ 5-S5	Medical Records	Total I 1.7%/ I-1.9%/ 2-29.0%/ 3-30.9%/ 4-30.0%/ 5-8.2% had swallowing disturbances
Han et al. [5]	127	69.1 ± 7.9	I-SI/ 2-S2/ 3-S3/ 4-S4/5-S5	Questionnaire	1-14.3%/ 2-16.7%/ 3- 33.3%/ 4- 50.0%/ 5- 100.0% had dysphagia
Kim et al. [12]	33	69.09 ± 1.42	\$1-\$5	VFSS	78.8% had penetration or aspiration
Michou et al. [16]	22	35-80	NA	VFSS and Qestionnaire	II had swallowing disturbances*
Monteiro et al. [15]	30	44-89	2.1 ± 0.80	VFSS	40.0% swallowing complaints
Ou et al. [7]	518	61.94 ± 10.67	2.0 ± 1.00	Questionnaire	36.3% had dysphagia
Perez-Lloret et al. [6]419	69 ± 10	2.0 ± 0.50	Questionnaire	18% had dysphagia
Rajaei et al. [13]	33	66.09 ± 9.4	2.06	VFSS	45.5% had penetration/aspiration
Silverman et al. [14]	68	71.3 F/ 68.1 M	2.4 ± 0.90	VFSS	52.9% swallowing concerns
Walker et al. [8]	75	75	2.72 ± 0.79	Questionnaire	32.0% had dysphagia

Table 2 Sum of more common type of dysphagia treatment in Parkinson's disease

Management of Dysphagia	Туре	Exercises	Results	Literature
Behavioral	Diet modifications	Thickened liquids	Delay in swallowing initiation	Luchesi et al. [47]
			Reduction of penetration and aspiration	Troche et al. [44]
	Postural adjustments	Chin-tuck	Reduction of penetration	Logemann et al. [48]
				Luchesi et al. [47]
	Airway closure maneuvers	Effortful swallow	Reduction of stasis	Felix et al. [46]
				Troche et al. [51]
Rehabilitation	Strengthening and range of motion exercises	Oropharyngeal and laryngeal musculature LSVT	Improving airway protection, cough function and tongue mobility	Luchesi et al. [47]
				Russel et al. [49]
				El Sharkawi et al. [50
	EMST	Strengthening of expiratory muscle	Improve pulmonary function and coughing	Kim et al. [52]
				Wheeler et al. [53]
	Oral stimulation	Taste	No evidences long term treatment	Butler et al. [56]
		Smell		
		Temperature		

Motors aspects of dysphagia in parkinson's disease

The dysphagia in PD is often associated with oral phases of swallowing because of the difficulty of bolus propulsion and tongue manipulation, slowness to initiate oral manipulation, presence of drooling and premature spillage, and presence of oral residue. Even in pharyngeal stage of swallowing abnormalities were found, such as, reduction in lower airway closure, delay in the initiation of pharyngeal phase of swallowing, residue in the valleculae or pyriform sinuses and on the aryepiglotic folds or posterior pharyngeal wall, reduction of the upward and forward motion of the hyoid that can results in abnormalities of upper esophageal sphincter opening. ¹⁸ The rigidity, hypertonia, bradykinesia and involuntary movements can interfere in

the motor control of swallowing, increasing the risk for penetration and laryngeal aspiration.

An important aspect to observe during clinical evaluation of swallowing is the respiratory-swallowing coordination. The respiratory pause duration through the swallowing and the expiration after swallowing are recognized as important aspects to airway protection. This coordination in PD patients can influence the severity of dysphagia.¹⁹ Troche et al.,¹⁹ observed that the most severe cases of laryngeal penetration and aspiration of liquids, in PD patients, had episodes of inspiration after swallowing and shorter swallowing respiratory pause durations. Another important aspect related with dysphagia and respiration in DP is the cough efficiency. Cough is an important mechanism of airway protection and usually the most common sign of laryngeal penetration/aspiration.²⁰ However, in PD patients, cough is frequently less elicited, even in early disease stage. 15 The strength and coordination to produce a sequential cough, is very important for cleaning lower airway. When considering patients with PD and dysphagia this ability is usually impaired in comparison with patients without dysphagia.21 Some studies evidenced an increased chance of penetration and/or aspiration when the cough reflex is not triggered or delayed.²² Additionally, in cases of moderate and severe dysphagia the triggered cough reflex showed less effectiveness when compared with voluntary cough.21

Non-motors aspects of dysphagia in parkinson's disease

In addition to the known motor alterations (tremor, bradykinesia, rigidity and hypertonia), the non motor symptoms as cognitive, somatosensory, smell, and taste symptoms can also negatively interfere on the swallowing function in PD patients. Cognitive aspects are very relevant to the control and planning of swallowing,²³ especially, regarding to volitional condition, manipulation, chewing and propulsion of the food in the preparatory and oral phases of swallowing. In a course of the neurodegenerative process of the PD is very common the appearance of dementia. Dysphagia was strongly associated with dementia in a long term study with 6462 patients with PD.¹¹ However, even without the diagnosis of dementia, PD patients can have problems due to lack of attention during meals. Brodsky et al.,²³ noted that PD patients can have the swallowing phases impaired if their attention is divided with other tasks. The factor attention when eating, it is very significant to guide the patient and their caregivers, warning to take care with several stimuli during the day-to-day eating environment.

Saliva control is an important aspect in PD, which has been intensively investigated in the last 5 years. Additionally of the difficulty to swallowing, these patients have a faster excretion of saliva, leading to easier saliva retention in the mouth or sialorrhea (drooling).^{24,25} Ou et al.,⁷ reported that 52.7% of the PD patients complained about sialorrhea, and this symptom was more frequent in patients with a lateonset of the disease. Also, Kalf et al.,²⁵ observed that the patients, in most advanced stages of PD, with sialorrhea attempted to compensate the excess of saliva increasing the swallowing frequency.

Oropharyngeal somatosensation, smell, and taste are the predominant sensory inputs transmitted from the oral periphery to the brain through separate but integrated neurophysiological pathways.²⁶ The sensory receptors known as chemoreceptors, mechanoreceptors, nociceptors, proprioceptors and thermoreceptors-exist in the mucosal lining of the oral, lingual, pharyngeal, laryngeal, and esophageal structures.²⁷ These receptors receive and carry modality-specific sensory information through afferent pathways to the brainstem and

the brain, where a sequence of sensor motor communications will lead to the oral and pharyngeal motor events (i.e., mastication, oral manipulation of the bolus, and ultimately swallowing).²⁶ Decrease or complete loss of olfactory function, called as hyposmia or anosmia, respectively, are symptoms present in approximately 90% in patients with PD and often precede motor symptoms.²⁸ The olfactory change is progressive, and are present in early disease stage^{28,29} There is also evidence that the sense of taste is affected during the PD progression, characterized by difficult to recognize specially the salty and bitter taste in PD patients.³⁰⁻³²

Smell and taste loss was perceived in a population study, with a frequency between 2 to 10 years before de perception of motor symptoms.³³ The exact relationship between loss of smell and taste and motor control of swallowing as well as airway protection is not completely understood. Authors assumed that both function, smell and taste identification are very important to the anticipatory phase of swallowing, the choice of the food, the pleasure of eating, and the motor planning of the swallowing process. Moreover, in patients with PD and Parkinsoni an syndrome, loss of smell and taste functions were associated with signs of bolus penetration in the lower airway.³⁴

A reduction of sensitivity can be present in the pharynx³⁵ as describe by Mu et al.,³⁵ The pharynx has an important role in the bolus conduction, and also, in triggering the swallowing reflex. Mu et al.,³⁵ found a large number of muscular fibers atrophied in the pharynx, an additionally alteration in sensorial nerves, such as: glossopharyngeal nerve, pharyngeal sensory branch of the vagus nerve and the internal superior laryngeal nerve. This sensorial alteration leads to an insufficient auto-perception of swallowing problems and several of PD patients do not complaint about dysphagia.

Thus, the treatment of Parkinson's disease needs to consider not only motor impairments, but also the sensorial aspects. Monteiro et al., 15 studied 35 subjects with PD and observed that 22% of the dysphagic patients with laryngeal penetration had no swallowing complaint. This is an alarming evidence if we consider that, many times, the physicians only recommends a swallowing treatment to a patient when they have complaints like choking while feeding. In PD patients this increased time until initiate a swallowing management can bring serious consequences, as pneumonia and malnutrition, including less life expectance. According to Akbar et al., 36 between 1979 and 2010 there was an increase of pneumonia cases in American PD patient compared to paired-age controls.

Management of dysphagia in PD

According to the American Speech-Language-Hearing Association (ASHA) the speech-language pathologist (SLP) is defined as the professional who engages in professional practice in the areas of communication and swallowing. The SLP is responsible for the evaluation and treatment of swallowing and communication difficulties. Regarding the swallowing problems, the SLP does the two most commons instrumental assessment, Fiber optic Endoscopic Evaluation of Swallowing (FEES) and Video fluoroscopy of Swallowing (VFS), and clinical evaluation such as beside assessment. In the clinical evaluation, as the patients swallowed, cervical auscultation was performed to add information about abnormal signs at the pharyngeal swallowing³⁷ phase. Oral bolus transit time, anterior or posterior escape, positive cervical auscultation (with signs that indicate a presence of stasis or penetration with aspiration risk), coughing (before, during, or after swallowing), and wet voice were also observed.

Based on the evaluations above the SLP plans the treatment. The intervention can comprehend several techniques to improve swallowing. According to Robbins et al.,³⁷ these techniques can be categorized according to its aim in compensatory, motor without the swallow act, motor with the swallow act and sensorial. In the "compensatory" category, the maneuvers used were chin-tuck to improve airway protection during swallowing and bolus consistency to facilitate the feeding of patients with decreased coordination of tongue, reduced contraction of pharynges, delay in triggering swallowing reflex, reduced airway protection, and chewing difficulty.

In the "motor without swallow act" category, the following were used: (i) strengthening-tongue to improve bolus propulsion; (ii) tongue control to improve tongue mobility and facilitate the bolus management in the oral cavity; (iii) shaker to increase strength in supra hyoid muscles, reducing the penetration and aspiration risk due to stasis in pyriform sinus; (iv) vocal exercises to improve airway protection through the improvement in the glottis adduction.

In the "motor with swallow act" category, the following were used: (i) effortful swallow to increase strength to eject the bolus and to approximate the larynx structures, improving airway protection; (ii) tongueholding (Masakomaneuver) to increase movements of pharyngeal muscles against the basis of the tongue during the act of swallowing; (iii) frequency of swallowing (multiple swallows) to clear stasis. The "sensorial" category includes bolus effects (changes in volume, viscosity, temperature, or taste in order) to improve oral and pharyngeal sensibility and control bolus management.

SLP intervention for dysphagia in PD is one of the most performed treatment in some countries around the world. The swallowing management in PD usually aims to improve the swallowing act and introduce compensations in order to provide a safer feeding. Some of more common type of dysphagia treatment in Parkinson's disease³⁸ is exposed in Table 2. Unfortunately, even in the recent times, many patients are not aware about the possibility of Speech-Language Therapy for communication and swallowing aspects. Miller et al.,³⁸ interviewed 215 PD patients and caregivers, and 43% of the interviewed had no contact with this type of therapy. Additionally, all the patients that underwent a SLP treatment referred to it as very positive. PD patients that were not treated by SLP professionals reported the difficult of access to this type of intervention and the need to be early directed to it by the physicians and other health professionals.

The benefits, of swallowing and communication treatment in PD, both in short and long term, have being reported by some studies in the last 5 years. 17.39-41 The results showed improvement in quality of life and swallowing functionality, and reduction of the residues after swallowing, among other benefits. Unfortunately, it is required further studies to confirm the effectiveness of many maneuvers and techniques used in the SLP swallowing therapy, and its effect in long term to maintain a safer feeding process. The literature has shown that the earlier treatment of swallowing can contributes to a safer oral feeding for a long time in these patients. Suntrup et al. 2 found an adaptive cerebral change even in patients with DP and dysphagia diagnostic, as a result of compensation to execute swallowing function. Luchesi et al., 17 observed a significant improvement in swallowing functionality after 10 months of SLP therapy in DP patients. Moreover, these patients frequently show an increase of quality of life after the SLP therapy.42,43

Dysphagia management includes compensatory measures to protect the airway (eg, postural adjustments, airway closure maneuvers, diet modifications) and behavioral swallowing rehabilitative treatments (eg, strengthening and range of motion exercises). Rehabilitation of sensor motor function, such as swallowing, after brain damage can occur best through experiences that modify the brain's structure and function (i.e, experience-dependent brain plasticity).³⁷ Given the high complexity of swallowing, the combination of exercises and maneuvers is the best option of treatment. Thus, the interventions consist in a great variability with respect to treatment combinations, frequency, and intensity according to the need of the patient. In PD patients, compensatory interventions are usually recommended to redirect the bolus away from the airway. An example is the use of thickened liquids, which exhibit an immediate reduction of penetration and aspiration.44 Therefore, to evoke a neural plasticity impact and consequently cerebral changes, it is necessary to apply a functional treatment of swallowing. Robbin et al., 37 discuss the importance to use maneuvers and exercises involving the swallowing act based on the 10 principles of neural plasticity ⁴⁵ use it or lose it, use it and improve it, plasticity is experience specific, repetition matters, intensity matters, time matters, salience matters, age matters, transference and inference.

The maneuver of effortful swallow offers a great muscular activity, it improves the bolus propulsion and approaches the larynx cartilages, increasing airway protection. This maneuver was designed to improve the posterior movement of the base of the tongue, which is stiff and slowed in PD patients,46 and prevent the accumulation of stasis in the vallecula, and had its effectiveness confirmed by Felix et al.,46 Luchesi et al.,47 in a long term study with 24 PD patients, found the head position change (chin-tuck), changing in bolus consistency and vocal exercises as strategies often recommended by SLP. Logemann et al., 48 showed evidences that in PD patients the chin-tuck maneuver combined with thick liquid can be important to prevent pneumonia, because of its immediately effect. Furthermore, vocal exercises can be able to improve the airway protection, cough function, and tongue mobility during the swallowing.⁴⁹ In the same way, the Lee Silverman voice Treatment (LSVT®) may influence oral and pharyngeal stage performance, particularly in terms of lingual biomechanics.⁵⁰

The may minimize the reducing airway closure. The EMST uses a valve to exercise the expiratory and sub mental muscles (important to coughing and swallowing). Studies with PD patients demonstrated improvement of swallowing functionality, reducing laryngeal penetration and aspiration, especially due to the better hyolaryngeal displacement during swallowing. 51-53 The behavioral swallowing rehabilitative treatments include the strengthening and range of motion exercises and treat of sensory impairments. Exercises for tongue strengthening (exercises aiming at propulsion of bolus) and control were frequently recommended due to tongue movements of PD patients.⁴⁷ The tongue movement in these patients, especially those responsible for bolus manipulation, chewing, and propulsion are considered bradykinetic/hypokinetic and weak in the oral phase^{54,55} Consequently, oral phase of swallowing is usually long and tongue pressure progressively reduced during the disease course. 55 However, shorter oral transit time not always is a good sign in PD. Argolo et al.,9 observed lingual pumping (repetitive, involuntary and anteroposterior movement of the tongue during the ejection of the bolus to the pharynx) associated with pharyngeal retention of food and laryngeal penetration in patients with a shorter oral transit time. The reduction of tongue strength, essential to propulsion of solid and pasty food, is yet expected in elders, but in PD it is worse due to nigrostriatal dopamine depletion⁵⁴ and an attention to this aspect is required during the therapy planning.

The assessment of non-motor aspects of dysphagia in PD, has become imperative to effectively treat sensory impairments. However, we do not have scientific evidences of the benefits of this approach ⁵⁶ thus, studies are reinforced. More recently, studies discuss the transcranial magnetic stimulation in cerebral cortex ¹⁶ neuromuscular electrical stimulation ³⁹ deep-brain-stimulations in subthalamics nucleus ^{57,58} and surface electrical stimulation in sub mental region. ⁵⁹ To date, none of these studies reported a significant improvement of swallowing with only one type of stimulation. In conclusion, the dysphagia in Parkinson disease is a common disorder that needs to be assessed and treated as soon as possible to contribute to better nutritional condition, hydration and quality of life. Intensive and specific treatment to the swallowing disorders reinforces the importance to have a clinical and also instrumental swallowing assessment.

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Conflicts of interest

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