



## Review Article

## Effective Swallowing Rehabilitation Strategies in Patients with Multiple Sclerosis Suffering from Dysphagia: A Literature Review

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### ARTICLE INFO

#### Article History:

Received: 29/02/2020

Revised: 23/08/2020

Accepted: 02/09/2020

#### Keywords:

Multiple sclerosis

Rehabilitation

Dysphagia

Deglutition

Please cite this article as:

Zinaee S, Rahmani S, Ghaemi H. Effective Swallowing Rehabilitation Strategies in Patients with Multiple Sclerosis Suffering from Dysphagia: A Literature Review. *JRSR*. 2020;7(3):106-113.

### ABSTRACT

**Background:** Multiple sclerosis (MS) is a disease of the central nervous system (CNS) which leads to various clinical signs and symptoms such as dysphagia. Given the prevalence and consequences of the disease, it is vital for patients with MS (PWMS) to be under medical supervision and receive specific care and services immediately. The present study aimed to comprehensively and inherently describe various Swallowing Rehabilitation Strategies (SRSs) and clarify the probable effects of each on the swallowing mechanism in PWMS suffering from dysphagia and to eventually answer the question: Which SRS(s) are effective on dysphagia in PWMS?

**Methods:** In this review of literature, search of the Web of Science, PubMed, and Science Direct databases was carried out for relevant studies published from 1990 to 2020 and written in English. The search resulted in 772 articles. Ultimately, 103 articles in total were selected for inclusion in the present study.

**Results:** The clinical results show that using different SRSs in dysphagia management can help reducing dysphagia and the risk for aspiration. These approaches are classified in four categories: compensatory methods, stimulation techniques, exercises, and maneuvers.

**Conclusion:** The current review reports the positive impact of SRSs such as electrical and sensory stimulation methods, maneuvers, and compensatory methods on dysphagia in PWMS. They can reduce the risk of aspiration for PWMS and, consequently, provide a better quality of life.

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### Introduction

Multiple sclerosis (MS) is an autoimmune, inflammatory, chronic, and degenerative disease of the myelin tissue (oligodendrocytes) of the central nervous system (CNS) [1]. Depending on the location of the lesion, various signs and symptoms may appear together in patient with MS (PWMS) [2]. The prevalence risk of MS is twice as high in females as in males, particularly females within the age range of 20 to 40 years [3]. Geographically, MS has the highest occurrence rates in northern Europe, North America, and Australia (0.05% to

2%) [4]. Conversely, Asia, Africa, and Central and South America have the lowest MS prevalence rates (0.01% to 0.03%) [4]. Eskandari et al. studied the prevalence of MS from 1991 to 2014 in Tehran and found it to be almost 0.1% percent. They also showed an increase in the prevalence of MS in both males and females during those years [5].

Clinical signs and symptoms differ among PWMS depending on the location of the CNS lesions [6]. PWMS may experience a variety of limitations (e.g., motor impairments, sensory impairments, cognitive deficits, visual deficits, mental impairments) as well as other issues such as pain, tremors, dysarthria, ataxia, apraxia, spasticity, depression, fatigue, sexual dysfunction, seizure, respiratory problems, dizziness, weakness, paralysis, or kidney and bladder dysfunction [6-13].

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In 1877, dysphagia was identified as one of the clinical signs of MS [14]. According to a lot of studies, almost one third of PWMS suffer from dysphagia [7, 14-24], and many studies have demonstrated the prevalence of dysphagia in PWMS [25]. Tarameshlu et al. have established that dysphagia is a common difficulty among Iranian PWMS [26]. Like other brain functions, the mechanism of swallowing is dominantly activated by a brain hemisphere; however, the brain activation is mostly bilateral in voluntary swallowing. Furthermore, there are areas in the forebrain, called the sensory-motor cortex and the premotor cortex, which are activated during chewing/swallowing and are in connection with the nucleus located in the corticobulbar fibers of the brainstem. The solitary nucleus and ambiguous nucleus are two other areas in the brain which play important roles during swallowing [23]. Hence, the probability of dysphagia in PWMS can be increased by damage to the dominant hemisphere (for swallowing), corticobulbar fibers, and especially the brainstem [18]. Moreover, the superior sensorimotor cortex of the brain is activated during tongue movements (voluntary stages of swallowing) [27]. The caudo-lateral-sensorimotor cortex of the brain is activated during the infusion of water into the oral cavity as well [27]. As the greatest force generator, the brainstem engages reflexively after the voluntary stages of swallowing [28].

The risk of dysphagia can be increased by increases in the severity of the disease [29]. Dysphagia can also be accompanied by impairments in mental behaviors, cerebellar functions, and the respiratory system [24]. Solaro et al., however, showed no significant correlation between patient's age and gender with the results of dysphagia on a multiple sclerosis patient-reported questionnaire which indicated dysphagia difficulties in PWMS [22].

According to the O'Neil dysphagia graduation scale, the severity of dysphagia is divided into three stages: mild, moderate, and severe [30]. In mild dysphagia, mild delays are seen in oral and pharyngeal phases. Patients have prolonged meals, and the food remains in the mouth and pharynx, but the patients can clean them spontaneously [30, 31]. Moderate dysphagia may begin immediately and be a consequence of the worsening of mild dysphagia [30, 31]. In this case, a backward movement of foods, occurrence of dysphagia during swallowing, fluid leakage into the airway causing choking, apnea, and aspiration may also be observed [30, 31]. Severe dysphagia is characterized by a lack of oral nourishment, the presence of a nasogastric tube, and/or gastrostomy feeding [30, 31]. In severe dysphagia, the foods may reside in the mouth and/or pharynx and cannot be cleaned by voluntary coughing, whereby silent aspiration can consequently be seen [30, 31].

In PWMS, dysphagia may occur in each swallowing phase, including the oral preparatory, oral propulsive, pharyngeal, and esophageal phases. Calcagno et al. and Poorjavad et al. reported that dysphagia often occurs during the pharyngeal phase in PWMS and can be characterized by insufficient glottal closure and weakness of soft-palate/larynx movements [18, 20]. In their manofluoroscopy study, De Pauw et al. described

the oral phase in all PWMS with dysphagia, which is characterized by a delay in the swallowing reflex onset and difficulties in bolus manipulation [32]. They also reported that the pharyngeal phase is accompanied by reduced contractions in the hypopharynx or limited opening of the upper esophageal sphincter in patients in severe stages [32]. According to their study, most PWMS who suffer from dysphagia also experience a change in their feeding habits [32]. Borucki et al. showed that the risk of dysphagia in the oral phase can be associated with cerebellar dysfunctions, and the risk of dysphagia during the pharyngeal phase can be associated with defects in brainstem function, mental disorders, and/or cerebellar disorders [33]. Abraham et al. established that abnormalities in cerebellar functions occur in all PWMS suffering from dysphagia [34].

The clinical signs and symptoms of dysphagia in PWMS usually include changes in feeding habits, coughing or choking during/after swallowing, drooling, nasal regurgitation, multiple swallowing, difficulty in initiating swallowing, recurrent pulmonary infections, and difficulty managing secretion [23, 32].

The risk of death due to pulmonary infections is 4.7 times higher among PWMS than in normal people [35]. Therefore, aspiration should be noted as a significantly dangerous consequence of dysphagia in PWMS, because it can increase the risk of mortality, especially in the late stages of the disease [7, 18, 23, 31]. Aspiration can also take place silently (known as silent aspiration) in 50% of patients with neurological diseases such as MS [36].

To date, only three review studies have been conducted to identify the effect of swallowing rehabilitation in PWMS [16, 19, 37]. First, Giusti and Giambuzzi investigated dysphagia management in PWMS as a review [19]. Their review abstractly studied dysphagia management divided into three branches, which were described according to the three stages of MS mentioned above [19]. Second, Alali et al. titled their review "Treatment Effects for Dysphagia in Adults with Multiple Sclerosis" [16]. In contrast to the title of their study, the authors mentioned within the methodology section that a comprehensive literature search was independently carried out by the authors to find articles which investigated the treatment effects for pharyngeal dysphagia in adults with MS [16]. This systematic review ultimately identified as eligible those studies which consisted of instrumental interventions and excluded behavioral intervention studies (e.g., muscle strengthening exercises or training in compensatory strategies for safer swallowing) [16]. According to the four phases of the swallowing mechanism as well as the title and methodology of this study, it seems that the research was unintentionally conducted erroneously, which probably resulted in mistakes that consequently provided paradoxical results. Third, D'Amico et al. also reviewed the literature to give an overview of the dysphagia phenomenon in PWMS in accordance with diagnostic and therapeutic methods. Furthermore, they described the importance of an early and integrated multidisciplinary approach to treatment [37]. This review also provided abstract insight related to its purpose [37]. Moreover, in contrast

to the title of this study (Management of dysphagia in multiple sclerosis; current best practice), the study did not introduce a current best practice relative to other practices [37]. Therefore, as those three review studies seem not to have investigated the literature appropriately and did not provide precise results, the present study was conducted to comprehensively and inherently describe various SRSs, to clarify the probable effects of each on the swallowing mechanism in PWMS suffering from dysphagia, and to eventually answer the question: Which SRSs are effective on dysphagia in PWMS?

## Methods

This literature review began with a search of the Web of Science, PubMed, and Science Direct databases to initially gather literature, especially grey literature. Those databases were chosen, because they are extensively utilized by scientific and healthcare communities.

Inclusion criteria for studies in this research were: (1) published between 1990 and 2020; (2) relevant to the aim of the present study; (3) written in English. Repetitive studies were excluded. Additionally, potential publication bias was considered by the authors in the present study. Therefore, the Critical Appraisal Skills Programme (CASP) diagnostic checklist was applied to evaluate the potential publication bias of the collected studies [38]. Each author independently completed each checklist to judge the quality of papers and then decide which study was to be deleted or kept in the process. If two authors decided to remove/keep a study, the decision was accepted. Furthermore, the authors decided to keep some papers with lower relevance to provide a more comprehensive insight into MS and/or SRSs.

## Results

The initial search identified 772 articles. Then their

titles and abstracts were carefully reviewed for eligibility in accordance with the inclusion criteria. Moreover, the authors immediately evaluated the articles to find any potential publication biases between them in accordance with the CASP diagnostic checklist. Minor bias was found in several studies. Consequently, 25 articles were selected as relevant and eligible, and 78 articles with lower relevance were chosen by the authors to supply a more comprehensive insight into MS and SRSs. Ultimately, 103 articles were selected from a total of 772 articles identified. The other 669 articles were excluded as a consequence of the search strategy. The search strategy is shown concretely in Figure 1.

To improve the health and quality of life of PWMS, treatment requires a specific, appropriate, accurate, and integrated approach as well as a multidisciplinary care team [39-42]. Fortunately, various rehabilitation approaches can be used for dysphagia management in PWMS; however, a specific drug treatment for MS-induced dysphagia has not yet been reported. Generally, SRSs can be classified into four categories: (a) compensatory methods, (b) stimulation methods, (c) swallow maneuvers, and (d) therapy techniques.

**1) Compensatory methods.** Initially introduced and applied by Larsen in 1972 [43, 44], these methods make the swallowing mechanism safer through a set of behaviors, but do not treat the cause of dysphagia [45]. However, they try to compensate for anatomical/functional insufficiency [46]. They make no changes in the swallowing pattern [47], but they change the manner of swallowing [48]. These strategies can provide a safer oropharyngo-oesophageal transition as well [49].

Calcagno et al. and Tarameshlu et al. showed that compensatory methods can reduce the risk of aspiration and be effective in the management of dysphagia in PWMS [18, 50]. The most common compensatory methods include (a) postural techniques; (b) food consistency modification; (c) bolus size modification.

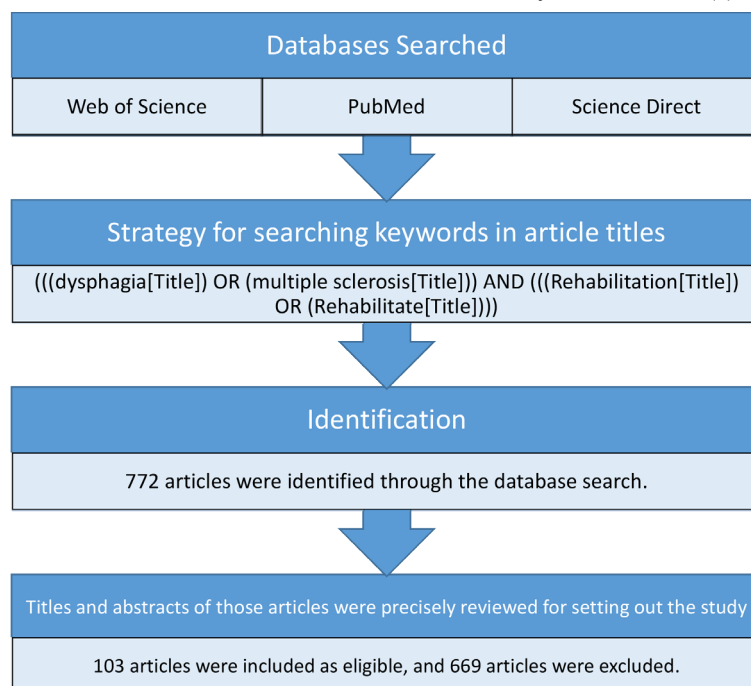


Figure 1: Search strategy diagram.

*Postural techniques* can modify the oropharyngeal tract and pharynx posture and facilitate bolus transition during swallowing [45]. Postural techniques should be applied before the oral intake and continued during oropharyngeal swallowing [45]. Moreover, they can be accomplished in combination or separately. These techniques are generally divided into the following six procedures. First is the lying down or one-side technique, which is commonly used in people who are unable to sit [51]. This procedure can also be applied for patients suffering from weakness of the base of the tongue [52]. Second, tilting the head forward (also known as chin down, chin tuck, or neck flexion) can be applied for patients suffering from a weakness of the base of tongue, oral bolus control problems, difficulty in pharyngeal swallowing initiation, insufficient vocal fold closure, or reduced laryngeal elevation [45, 53, 54]. This procedure increases control of the bolus in the oral cavity, extends the valleculae, permits the epiglottis to cover the airway entrance, strengthens movements of the base of the tongue, narrows the airway entrance, and decreases the risk of penetration or aspiration [45, 53, 54]. Third, tilting the head backward can be applied to compensate for defects or weaknesses in oropharyngeal transition [45, 54]. It must be noted that this procedure is not appropriate for patients who are at the risk of aspiration or penetration [45, 54]. In this procedure, the patient is asked to tilt his or her head backward after chewing and swallow immediately; then the bolus is passed into the pharynx by the force of gravity [45, 54]. Fourth, rotating the head to the damaged side facilitates glottal closure in patients with unilateral laryngeal paresis [54, 55]. This procedure closes the pyriform sinus on the injured side, increases the entrance of the upper esophageal sphincter, and facilitates the bolus entering the esophagus [54, 55]. The videofluoroscopic evidence shows the arytenoid compensatory movements in the healthy side during this procedure [55]. Fifth, rotating the head to the stronger side facilitates the bolus transition by the stronger side in people with one-sided weakness of the tongue or pharynx.

*Food consistency modification* has been more noticed recently as the main technique among compensatory methods [56]. Most PWMS complain of swallowing problems while drinking liquids in the moderate stage of the disease [32]. Furthermore, PWMS may suffer from swallowing problems while eating food in the severe stage of the disease [32]. Hence, food consistency modification and postural techniques can decrease the risk of aspiration or penetration [18]. Finestone and Greene generally divided food consistency into the two main classes of foods (e.g., pureed foods, minced foods, and soft foods) and liquids (e.g., pudding-like, honey-like, nectar-like, and thin) [57]. Bolus consistency can be modified to reduce the risk of aspiration or penetration [58].

*Bolus size modification*, based on the abilities of the patient, decreases the risk of aspiration and penetration and makes swallowing safer [59]. In some patients, using large boluses can result in more optimal swallowing [60, 61]. In contrast, some other patients optimally benefit from eating small boluses [62].

**2) Stimulation methods.** Stimulation methods can

generally be divided into two classes. The first is *sensory stimulation*, which lead to better initiation of swallowing reflex and esophagus peristalsis [52]. The second is *electrical stimulation* which is divided into the two procedures of (a) *electrical stimulation* of the neural tissues, and (b) electrical stimulation of muscle tissues. Electrical stimulation of the neural tissues may be provided to modify the function (inhibition or stimulation) of a nerve or a specific neural tissue [63]. Marrosu et al. showed that stimulation of the vagus nerve as a complementary tool can be useful in improving dysphagia in PWMS [64]. *Electrical stimulation* of neural tissues can also be applied for the cortex [56, 63, 65-68]. Electrical stimulation of muscle tissues can be used for one or more muscles [54, 69, 70]. Restivo et al. revealed in their pilot study that electrical stimulation of the pharynx was useful in managing dysphagia in PWMS [71]. Bogaardt et al. reported that electrical stimulation can improve the swallowing abilities of patients and decrease the risk of aspiration as well [72]. Moreover, Cosentino et al. showed that electrical stimulation of the swallowing motor cortex via transcranial direct current stimulation can improve the swallowing function of PWMS who suffer from dysphagia [73]. Restivo et al. also demonstrated in their recent pilot study that electrical stimulation of the pharyngeal motor cortex using transcranial direct current stimulation can significantly and advantageously increase swallowing abilities in PWMS and, consequently, reduce the risk of aspiration and penetration [74].

**3) Swallow maneuvers.** In swallowing rehabilitation, maneuvers have been devised to provide safe voluntary swallowing [53]. The maneuvers are inherently complicated for most patients to learn and thus requires a lot of practice [45].

The *Mendelsohn Maneuver* (MAN) facilitates the opening of the upper esophageal sphincter in patients who suffer from disruption of the opening (delay, reduction, and/or incoordination) [60, 75-79]. MAN is accompanied by a long swallow and brings the arytenoid cartilage and the hyoid bone closer to each other [80]. This maneuver also helps the opening of the upper esophageal sphincter via larynx elevation [49, 75, 76, 78, 81-83]. MAN improves the activity of the suprahyoid muscles as well [84].

The *Masako maneuver* (MAS) was designed to strengthen the obstruction of the pharyngeal cavity [45, 80, 85]. MAS also improves weakness of the base of the tongue and the posterior wall of the pharynx [54, 56, 86].

*Effortful swallow* (ES) improves the muscle contractions of the base of the tongue and prevents food from remaining in the valleculae [45, 49, 56, 86]. ES can increase the constriction between the tongue and the palate [87] as well as between the tongue and pharyngeal wall [88]. According to studies conducted on healthy people, ES increases intraoral pressure, duration of false vocal fold closure, laryngeal elevation, duration of pharyngeal constriction, and duration of rest in the upper esophageal sphincter, and it reduces the amount of residual food in the mouth [46, 54, 56, 86]. This method facilitates the peristalsis of the distal smooth muscle



region of the esophagus as well [89].

*Supraglottic swallow* (SS) was designed to improve the voluntary closure of the vocal folds, especially in patients suffering from insufficient closure before or during swallowing [45, 46, 54, 90].

*Super-supraglottic swallow* (SSS) is similar to SS and helps arytenoids move ahead and become close to the epiglottis [45, 54, 70]. In SSS, a sufficient closure occurs between the false vocal folds, whereby the airway entrance is closed [45, 54, 70]. The aim of this method is to improve the voluntary closure before or during swallowing and decrease the risk of aspiration [54]. Video-fluoroscopic and video-endoscopic evidence has shown that SS and SSS make the laryngeal closure even longer than the closure of normal individuals [91].

*McNeil protocol* (MP) is a relatively new protocol for managing dysphagia in adults [92]. MP aims to modify the weakness and incoordination of swallowing by focusing on effortful and rapid swallowing [92]. This therapeutic protocol seems to be similar to ES; however, MP has a hierarchy and is considered to be a systematic therapeutic approach [92]. Sia et al. and Crary et al. indicated that MP has a positive effect on larynx elevation in patients suffering from dysphagia [93, 94]. This protocol, however, has not yet been evaluated in any large control groups [93, 94].

Tarameshlu et al. showed that the use of MAN, MAS, ES, SS, and SSS can reduce the risk of penetration, aspiration, and the extent of residual food in the pharynx [50].

**4) Therapy techniques.** Various exercises have been designed as therapeutic techniques to improve swallowing ability [45, 53, 90, 95, 96]. They generally aim to improve the range, power, rate, and coordination of the movements or contractions of the orofacial, pharyngeal, and laryngeal muscles [45, 53, 90, 95, 96]. Moreover, they help modify muscle tone and facilitate the swallowing reflex initiation [45, 53, 90, 95, 96]. They can even incredibly affect the nervous system [46, 97]. The exercises should be applied in accordance with the swallowing problems [98]. Furthermore, it is better to use the exercises immediately after sensory stimulation when stimulation methods are used [45]. Tarameshlu et al. illustrated that using these exercises can improve swallowing ability in PWMS [50]. These techniques include the following exercises.

Lip exercises are designed to enhance the muscle tone of the lips [45].

Jaw exercises can be used to reduce the truisms (limitations in jaw opening) [54]. The patients should do these exercises alone if they could [54]. Sometimes, certainly, the patients should be supported unless they enable to do the exercises on their own [45, 99].

Tongue exercises have been devised to actively strengthen anterior-posterior movement, elevation, lateralization, and cupping of the tongue [54]. Pauloski explored whether active motor exercises of the base of the tongue can strengthen oral tongue movements and movements of the base of the tongue [54]. Tongue endurance exercises strengthen the weak tongue and improve the swallowing ability [100].

Bolus control exercises are designed to coordinate tongue

movements, chewing, and bolus manipulation [54].

Laryngeal exercises improve laryngeal movements during swallowing. Laryngeal elevation provides the upward movement of the larynx and closure of the airway; horizontal movements of the larynx help the opening of the upper esophageal sphincter [101, 102]. These exercises generally include falsetto, Shaker, and vocal fold adduction exercises. The *falsetto exercise* is a useful technique for patients suffering weakness in their larynx elevation [54]. The *Shaker exercise* is a worthwhile technique for patients who have dysfunction of the upper esophageal sphincter [103, 104]. This exercise improves the activation of the supra-hyoid muscles, laryngeal elevation, and opening of the upper esophageal sphincter [103, 104]. Finally, *vocal fold adduction exercises* are designed for patients suffering insufficient laryngeal closure [80]. Table 1 summarizes all of the methods discussed in this study.

**Table 1:** Swallowing rehabilitation strategies

Compensatory methods	Postural techniques
	Food consistency modification
	Bolus size modification
Stimulation methods	Sensory stimulation
	Electrical stimulation
Swallow maneuvers	Mendelsohn <i>Maneuver</i>
	Masako maneuver
	Effortful swallow
	Supraglottic swallow
	Super-supraglottic swallow
	McNeil protocol
Therapy techniques	Lip exercises
	Jaw exercises
	Tongue exercises
	Bolus control exercises
	Laryngeal exercises

## Discussion

The current study has shown that MS as an autoimmune disease can be associated with demyelination of nerve cells in the CNS. Moreover, this disease can cause a lot of physiological problems and have various manifestations, like dysphagia. One of the most dangerous and disadvantageous concerns for PWMS with dysphagia is recurrent pulmonary infections caused by aspiration which affect the quality of life and increase the risk of mortality. Thus, the presence, severity, and duration of dysphagia can seriously threaten the health of PWMS. Hence, a speech and language pathologist (SLP) as a member of the MS care team plays a particularly important role in assessing suitably, diagnosing early, and reducing dysphagia manifestations in PWMS [37].

The effects of various types of SRS on dysphagia have not been clarified in PWMS [32]. Likewise, no study has investigated the effects of methods such as the McNeill protocol and electrical stimulation of the brain cortex on dysphagia in PWMS. Marrosu et al., however, showed that using vagus nerve stimulation in PWMS may be more useful for improving swallowing ability when it is combined with some other swallowing rehabilitation strategies [64]. Bogaardt et al. also reported that electrical stimulation of the muscles between the chin

and the hyoid bone as well as the muscles between the hyoid bone and the thyroid cartilage can improve the swallowing ability and decrease the risk of aspiration in PWMS [72]. Furthermore, Restivo et al. demonstrated in their pilot study that pharyngeal electrical stimulation has a positive effect on dysphagia in PWMS [71]. Cosentino et al. showed that electrical stimulation of the swallowing motor cortex via transcranial direct current stimulation can improve the swallowing function of PWMS who suffer from dysphagia [73]. Furthermore, Restivo et al. recently demonstrated that electrical stimulation of the pharyngeal motor cortex using transcranial direct current stimulation can improve swallowing abilities in PWMS and, consequently, decrease the risk of aspiration and/or penetration [74]. Tarameshlu et al. showed that using MAN, MAS, ES, SS, SSS, compensatory methods, and sensory stimulation has advantageous effects on dysphagia in PWMS [50]. They also showed that such methods decrease the risk of penetration and aspiration in PWMS as well [50].

## Conclusion

Electrical and sensory stimulation methods, MAN, MAS, ES, SS, SSS, and compensatory methods can advantageously improve swallowing ability in PWMS suffering from dysphagia and, consequently, decrease the risk of aspiration as well as penetration. It is necessary to provide team-based healthcare services to help improve the quality of life and longevity in PWMS in accordance with the occurrence of MS as well as the prevalence of dysphagia and its consequences. Therefore, SLPs should precisely diagnose swallowing problems in patients suffering from dysphagia. In addition, SLPs must be aware and decrease the risk of aspiration early so as to reduce pulmonary infections as potential causes of death. It is also particularly crucial to properly apply the proposed SRS depending on the severity of the problems and clinical/paraclinical findings. Eventually, SLPs must accurately determine the outcome of the strategies before, during, and after the management by using clinical or paraclinical evaluations (like videofluoroscopic study) and rationally alter the management strategies if they are not effective.

## Acknowledgment

The authors express their appreciation to all researchers who sent us their papers or gave informative guidance and helped in the performance of this review. The authors declare that they have no conflicts of interest.

**Conflict of Interests:** None declared.

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