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New approaches to evaluation and management of dysphagia in neurological disease

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New approaches to evaluation and management of dysphagia in neurological disease

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Editorial: New approaches to evaluation and management of dysphagia in neurological disease

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Editorial on the Research Topic
New approaches to evaluation and management of dysphagia in neurological disease

Neurological diseases frequently cause dysphagia and consequent aspiration pneumonia, significantly impacting patient mortality, morbidity, and healthcare costs. It is therefore essential to implement better strategies for evaluating, addressing, and rehabilitating dysphagia. Several protocols and tools have been developed for dysphagia assessment, with videofluoroscopy and videoendoscopy being considered the gold standards (1). Moreover, recent advancements offer promising non-invasive and simple procedures and instruments for dysphagia assessment (2, 3). Effective rehabilitation protocols have also been established for clinical practice (4). Novel techniques, such as transcutaneous electrical stimulation of the neck, are being introduced and are expected to improve neuromuscular function (5, 6).

The utility of novel dysphagia assessment tools has recently been reported (7, 8). Nakamori, Ishikawa et al. investigated the use of electronic stethoscopes and artificial intelligence (AI) analysis for swallowing sound evaluation in patients with amyotrophic lateral sclerosis, suggesting potential applications in telemedicine and home care. Among these advancements, tongue pressure has gained significant attention due to its practical value. Fukuoka et al.'s minireview summarized the detailed relationship and limitations of tongue pressure and dysphagia in patients with Parkinson's disease (PD). Physiological functional imaging also offers new possibilities for evaluating dysphagia. Gu et al. focused on cortical compensation mechanisms in dysphagia following medullary infarction. Using blood-oxygen-level dependent functional magnetic resonance imaging, they observed increased activation in specific brain regions, including the bilateral precentral gyrus, postcentral gyrus, insula, thalamus, and supplementary motor areas, after rehabilitation. Notably, their study showed that effortful swallowing activated more brain regions than regular saliva swallowing, suggesting its effectiveness as an approach for rehabilitation. As previously mentioned, gold-standard evaluations are crucial, especially with a serious condition such as dysphagia. Cui et al. emphasized the importance of videofluoroscopic swallowing studies (VFSS) in dysphagia assessment, demonstrating significant differences in swallowing parameters between patients with PD and those with post-cerebral hemorrhage or infarction. They assert that standardizing quantitative VFSS parameters can improve the accuracy and consistency of dysphagia evaluation. Beyond evaluating for dysphagia, determining patients at risk of dysphagia is equally important in clinical practice. Karunaratne et al. reported that older individuals and patients with neurological disorders are more susceptible to developing or experiencing worsened complications, underscoring the need for proactive identification. Furthermore, a retrospective study by Choi et al. identified clinical predictors of dysphagia in patients with traumatic and non-traumatic cervical spinal cord injuries, focusing on forced vital capacity and tracheostomy as key predictors.

Several novel interventions on rehabilitation have emerged. Nakamori, Toko et al. investigated the effects of cervical percutaneous interferential current stimulation on dysphagia in patients with PD, reporting a significant improvement in oral cavity residue. Another study by Yeo et al. reported that therapeutic singing exercises may help maintain swallowing function and potentially improve swallowing-related quality of life in patients with advanced PD. The utility of acupuncture for dysphagia treatment has also been widely reported. Wu Y. et al. explored the combined use of postural control and electroacupuncture compared to conventional rehabilitation in 138 patients with post-stroke dysphagia. This integrative approach significantly improved swallowing function and reduced aspiration-related complications in the observation group than in the control group. This is further evidenced by the results of their standardized swallowing assessment and water swallowing test. Similarly, a systematic meta-analysis by Li et al. demonstrated the effectiveness and safety of acupuncture in treating aspiration among 1,284 patients with post-stroke dysphagia. Their findings revealed that acupuncture, used alone or combined with other therapies, significantly improved the Penetration Aspiration Scale scores, VFSS results, and hyoid bone displacement in these patients. Additionally, acupuncture showed a higher overall efficacy rate and fewer adverse events compared to conventional rehabilitation. In another study by Wu M. et al., data mining techniques were employed to analyze optimal acupuncture parameters for post-stroke dysphagia in 39 studies, which included needle size, retention time, treatment frequency, and core acupoints.

Comprehensive reviews on dysphagia and its guidelines have also been conducted. Bibliometric studies by Ye et al. and Guo et al. highlighted the evolving research trends in the study of dysphagia and stroke, such as aspiration, gastroesophageal reflux disease, neuroplasticity, and non-invasive brain stimulation. Likewise, He et al. utilized bibliometrics to identify research hotspots and cutting-edge trends in the field of post-stroke dysphagia rehabilitation. In their study, the keywords "validity" and "non-invasive brain stimulation" emerged as the most significant words in post-stroke dysphagia rehabilitation research. Regarding guideline studies, Gao et al. critically appraised clinical practice guidelines for managing dysphagia after acute stroke, revealing gaps between evidence and practice, particularly in applicability and stakeholder involvement.

The evaluation and management of dysphagia in neurological diseases have progressed significantly due to innovative therapeutic approaches, improved diagnostic techniques, and comprehensive research. Integrating these new insights into clinical practice can enhance patient outcomes, reduce complications, and improve quality of life. Continued research and collaboration among healthcare professionals are essential to further refine these strategies and develop effective personalized treatment plans for patients with dysphagia.

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Swallowing sound evaluation using an electronic stethoscope and artificial intelligence analysis for patients with amyotrophic lateral sclerosis

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Background and purpose: Non-invasive, simple, and repetitive swallowing evaluation is required to prevent aspiration pneumonia in neurological care. We investigated the usefulness of swallowing sound evaluation in patients with amyotrophic lateral sclerosis (ALS) using our new electronic stethoscope artificial intelligence (AI) analysis tool.

Methods: We studied patients with ALS who provided written informed consent. We used an electronic stethoscope, placed a Bluetooth-enabled electronic stethoscope on the upper end of the sternum, performed a 3-mL water swallow three times, and remotely identified the intermittent sound components of the water flow caused at that time by AI, with the maximum value as the swallowing sound index. We examined the correlation between the swallowing sound index and patient background, including swallowing-related parameters.

Results: We evaluated 24 patients with ALS (age 64.0 ± 11.8 years, 13 women, median duration of illness 17.5 months). The median ALS Functional Rating Scale-Revised (ALSFRS-R) score was 41 (minimum 18, maximum 47). In all cases, the mean swallowing sound index was 0.209 ± 0.088. A multivariate analysis showed that a decrease in the swallowing sound index was significantly associated with a low ALSFRS-R score, an ALSFRS-R bulbar symptom score, % vital capacity, tongue pressure, a Mann Assessment of Swallowing Ability (MASA) score, and a MASA pharyngeal phase-related score.

Conclusion: Swallowing sound evaluation using an electronic stethoscope AI analysis showed a correlation with existing indicators in swallowing evaluation in ALS and suggested its usefulness as a new method. This is expected to be a useful examination method in home and remote medical care.

KEYWORDS

electronic stethoscope, artificial intelligence, swallowing sound index, dysphagia, amyotrophic lateral sclerosis

Introduction

Amyotrophic lateral sclerosis (ALS) is a neurodegenerative disease mainly affecting motor neurons. It causes progressive weakness, muscular atrophy, dysarthria, dysphagia, and dyspnea. Currently, there is no disease-modifying therapy available; therefore, it is important to manage nutrition, respiration, and communication. Swallowing dysfunction caused by bulbar dysfunction is a major factor in determining patient prognosis (1, 2). Aspiration pneumonia, caused by swallowing dysfunction, is a critical problem. Aspiration pneumonia is associated with high mortality rates in patients with ALS (3). The evaluation of swallowing function is important for assessing the risk of aspiration and monitoring the progression of neurological impairment. Videofluoroscopic or endoscopic examination is the gold-standard method for evaluating swallowing function. There is not much burden on the patient during the examination; however, limited facilities use these types of equipment. Therefore, a non-invasive screening method for evaluating swallowing dysfunction should be developed.

Simple and non-invasive instruments have been developed to evaluate the swallowing function. Tongue pressure measurement prevails (4–7). In addition, the usefulness of tongue ultrasonography for measuring tongue thickness and motion has been reported (7, 8). However, these methods primarily reflect only the oral phase of swallowing.

With the exception of videofluoroscopic or endoscopic examinations, there are few established methods for the evaluation of the pharyngeal phase (9). Recently, the evaluation of swallowing sounds, including deep learning analysis, has become the focus, and several types of instruments have been developed (10, 11). Especially, considering the anatomical aspects of swallowing sounds and their actual swallowing and reproducibility, commercialized and implementable tools have been introduced in the medical field. These methods have been used to predict the risk of aspiration and to assist in the selection of special diets provided to patients (11). We have combined electronic stethoscope techniques and artificial intelligence (AI) to assemble a system that incorporates sound recognition and swallowing sound quantification. In particular, the quantification of swallowing sounds using deep learning techniques provides objectivity and is a differentiating feature from traditional stethoscopes. Additionally, these systems do not necessarily require direct auscultation by medical staff as patients or caregivers themselves touch their own bodies, which makes remote medical checks possible. During a pandemic, these types of assessments are extremely useful and important.

This study aimed to investigate the usefulness of an electronic stethoscope for evaluating swallowing dysfunction in patients with ALS by comparing several disease parameters and Indicators of swallowing function.

Materials and methods

Standard protocol approvals, registrations, and patient consents

The study protocol was approved by the ethics committee of Hiroshima University Hospital (E-1599-1) and was in accordance with the guidelines of the 1964 Declaration of Helsinki. Written informed consent was obtained from all the patients or their relatives. All the data analyses were performed in a blinded manner.

Participants

Consecutive patients with ALS who were diagnosed with definite, probable laboratory-supported ALS according to the revised El Escorial criteria (12) and admitted to Hiroshima University Hospital between 1 November 2021 and 31 December 2022 were enrolled in this prospective study. We excluded patients who could not tolerate full oral intake because of the risk of aspiration. Additionally, the swallowing sound index was evaluated using an electronic stethoscope and AI analysis of 57 healthy young volunteers.

Swallowing test protocol

The swallowing sounds were recorded using an electronic stethoscope in a silent room. To exclude the effect of noise during the test, patients were given non-verbal instructions. The gestures were shown alone, and the patients were instructed to drink water. Each participant was seated on a chair with the back vertically fixed at 90°. Participants were instructed to drink water while sitting. The patients drank 3 mL of water. This was adopted based on the revised water swallowing test, one of the most commonly used screening methods for swallowing evaluation, which uses 3 mL of water. The water was at room temperature and measured with a 5-mL syringe (ss-20ESzp, Terumo, Tokyo, Japan), which was injected into the patient's mouth, after which a gesture was made for the patient to start swallowing.

Swallowing sounds were recorded in the 2 Hz to 20 kHz wavelength band using an electronic stethoscope (MSS-U10C; Pioneer, Tokyo, Japan) placed at the top of the sternum below the sternal notch. After exploring various sites for attaching the stethoscope such as the neck and chest, it was determined that the top of the sternum below the sternal notch, where motion artifacts are least likely to occur, is the most appropriate choice. The sound data were transferred to a waveform audio file format via Bluetooth, and the collected data were analyzed using a dedicated AI application.

Swallowing sound evaluation using AI

In this study, a fine crackle sound discrimination AI algorithm for alveolar sound analysis was used to evaluate swallowing sounds, as previously reported (13). We have created algorithms not only for fine crackles but also for coarse crackles, wheeze, and rhonchi. Upon comparing and analyzing these sounds, we determined that the algorithm for fine crackles is the optimal choice. The AI analysis algorithm for calculating fine crackle sounds included 50 labeled sounds comprising characteristic frequency bands/continuations as teacher data for machine learning. Specifically, feature parameters (x) were extracted from the fine crackle sound caused by the inflow of water into the esophagus during swallowing using frequency, local variance, cepstrum analysis, the liftering process, and other methods. Next, the coefficients (a) of the feature parameters (x) were derived using AdaBoost as a machine-learning algorithm, and 50 pieces of labeled teacher data were added. The feature (y) was calculated using the following equation, consisting of the feature parameters (x) in the interval to be auscultated, and the coefficients (a) were determined using machine learning:

$$y = \sum_{i=1}^{148} a_i x_i,$$

a: Feature parameter coefficients calculated using machine learning

x : Feature parameter (normalized to $-1 \leqq x \leqq 1$).

The features (y) calculated for each frame at 12-ms intervals were converted to water inflow sound presence/absence data for each frame by comparison with a threshold determined using machine learning. The quantitative value of the fine crackle component (FCQV) was calculated for each second, based on the total number of frames and the number of frames in which the fine crackle sound was present.

$$INDEX = \frac{No.\ of\ target\ sound\ frames}{Total\ frames\ in\ auscultation\ section} \times 100$$

In the present study, we used FCQV to perform acoustic analysis of water inflow sounds. When evaluating the analysis algorithm, a discriminator was created that discerned whether the target sound existed if the FCQV exceeded a certain value (Figure 1).

The electronic stethoscope had a contact-type pressure sensor in the diaphragm to switch it on and off at the start and end of the auscultation, respectively. Our system excluded 0.2 s immediately before and after the auscultation from the target auscultation section.

Sample size

We calculated the required sample size based on a previous investigation of interstitial lung disease using an electronic stethoscope and AI analysis published by our research group (14). The minimum difference and standard deviation that we considered were 0.08 and 0.06, respectively. Based on an alpha level of 0.05 and a power of 0.80, we estimated that we would require a total of 20 participants.

Data acquisition

The participants were functionally rated by neurologists using the ALS Functional Rating Scale-Revised (ALSFRS-R) (15). The ALSFRS-R is a validated questionnaire-based scale that measures physical function and performance of activities of daily living. The total possible score is 48, and lower scores correlate with increased disability. The ALSFRS-R is divided into five domains: bulbar-related (speech, salivation, and swallowing), upper limb-related (dressing and hygiene, turning in bed, and adjusting bed clothes), lower limb-related (walking and climbing stairs), and respiration-related (dyspnea, orthopnea, and respiratory insufficiency).

Tongue pressure was measured using a tongue pressure manometer equipped with a balloon probe (TPM-01; JMS Co. Ltd., Hiroshima, Japan). To measure the tongue pressure, the patients were asked to hold the cylinder so that the balloon could be placed between the tongue and the anterior part of the palate with the lips closed. Each subject was then asked to compress the balloon onto the palate for 7 s, three times at 1 min intervals. Measurements were performed as previously described (4, 16). The reliability of intraindividual measurements has been previously reported (17). The maximum value among the three measurements was considered as the tongue pressure for each patient.

We also performed a comprehensive swallowing evaluation using the Mann Assessment of Swallowing Ability (MASA) score. The MASA score is an established, concise, and comprehensive assessment tool that indicates the risk of swallowing dysfunction in patients with stroke (18). The MASA consists of 24 items with a total potential score of 200. It has also proven useful for various other diseases and types of patients with dysphagia (19). Furthermore, among the 24 items, pharyngeal phase-related eight items (gag, palate, cough reflex, voluntary cough, voice, tracheostomy, pharyngeal phase, and pharyngeal response), which account for 70 points, were evaluated.

These measurements were performed at the same time as electronic stethoscope recording.

Statistical analysis

The data are expressed as mean ± standard deviation or median (minimum, maximum) for continuous variables and frequencies and percentages for discrete variables. Statistical analysis was performed using JMP 16 statistical software (SAS Institute Inc., Cary, NC, USA). The statistical significance of the intergroup differences was assessed using *t*-tests or χ^2 tests, as appropriate. Baseline data of patients with ALS were analyzed, and two-step strategies were employed to assess the relative importance of the variables in their association with the swallowing sound index using multiple logistic analysis. First, a univariate analysis was performed. Subsequently, a multi-factorial analysis was performed with selected factors that had a p-value of < 0.05 in the univariate analysis and age. A p-value of < 0.05 was considered to be statistically significant.

Results

During the study period, a total of 24 consecutive patients with ALS were investigated. Table 1 displays the background and physical characteristics of patients with ALS. The swallowing sound index measured and calculated using the same method as this study for 57 healthy young individuals (mean age 24.4 ± 1.9 years, 21 women, 36 men) was 0.369 ± 0.111, which was significantly higher than that of the patients with ALS ($p < 0.001$). The characteristics of the healthy young individuals are shown in Supplementary Table 1. Among the healthy young individuals, the mean swallowing sound index of men tended to be higher than that of women ($p = 0.06$); however, tongue pressure and body mass index were not correlated with the swallowing sound index ($p = 0.84$ and 0.58, respectively).

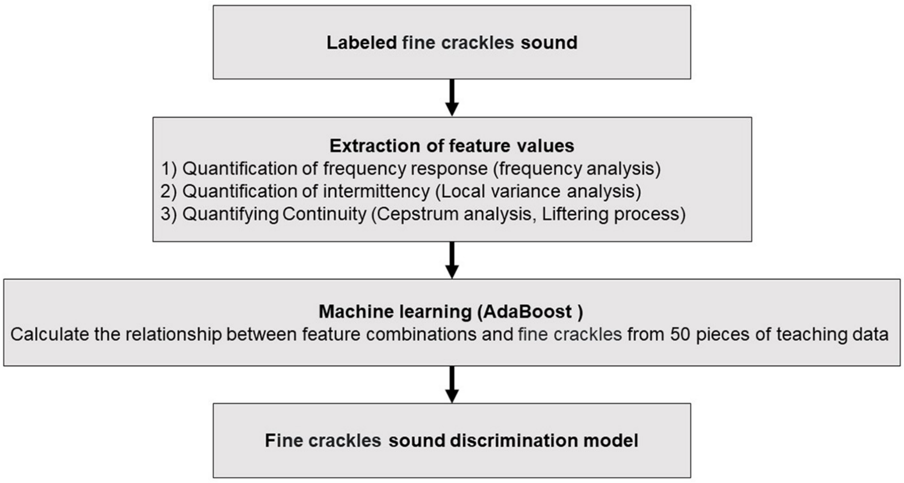

FIGURE 1
Algorithm for calculating swallowing sound index. The procedure for the swallowing sound analysis starts with the identification of fine crackle sounds, which are then subjected to feature value extraction through various techniques including frequency, local variance, and cepstrum analyses. These extracted features are fed into an AdaBoost machine-learning model, which is trained on 50 pieces of teaching data to discern the relationship between feature combinations and fine crackles.

TABLE 1 Patients' background.

Factors	$N = 24$
Age, year	64.0 ± 11.8
Sex (female), n (%)	13 (54.2)
Body mass index, kg/m^2	21.9 ± 3.3
Duration from onset, month, median (minimum, maximum)	17.5 (4, 114)
ALSFRS-R score, median (minimum, maximum)	41 (18, 47)
ALSFRS-R bulbar symptom score, median (minimum, maximum)	11.5 (4,12)
Onset type	
Limb, n (%)	19 (79.2)
Bulbar palsy, n (%)	5 (20.8)
Serum albumin, g/dL	4.1 ± 0.4
% vital capacity	80.5 ± 20.4
Tongue pressure, kPa	27.6 ± 16.2
MASA score	195 (151, 200)
MASA pharyngeal phase-related score	70 (57, 70)
Swallowing sound index	0.209 ± 0.088

ALSFRS-R, Amyotrophic Lateral Sclerosis Functional Rating Scale-Revised; MASA, Mann Assessment of Swallowing Ability.

The number of patients who complained of dysphagia was 11. Comparing the groups of the ALSFRS-R bulbar symptom score with a full score ($n = 12$) and <12 ($n = 12$), the swallowing sound index of the group with a bulbar sub-score <12 was significantly lower (Figure 2A). Scatter plots of the ALSFRS-R bulbar symptom score and the swallowing sound index are shown in Figure 2B. A positive correlation was observed between the ALSFRS-R bulbar symptom score and the swallowing sound index, indicating a decrease in swallowing sounds with the progression of ALS.

To explore the factors related to the swallowing sound index, a univariate analysis was performed on the factors listed in Table 1, revealing significant correlations with the ALSFRS-R score, the bulbar symptom score of ALSFRS-R, percentage of vital capacity (%VC), tongue pressure, and MASA ($p < 0.05$). Multivariate analyses were performed using individual factors and age. The results also revealed that the ALSFRS-R score, the bulbar symptom score of the ALSFRS-R, %VC, tongue pressure, the MASA score, and the MASA pharyngeal phase-related score were independently associated with the swallowing sound index (Table 2).

Receiver operating characteristic (ROC) analysis was used to determine the cutoff value of the swallowing sound index that defined ALSFRS-R <12, and a result of 0.228 was obtained (area under the curve =0.792, sensitivity 66.7%, specificity 83.3%, and $p = 0.005$).

Discussion

In this study, we investigated the utility of a novel non-invasive and easily repeatable swallowing evaluation method, utilizing AI analysis of swallowing sounds via an electronic stethoscope, for patients with ALS.

Videofluoroscopic or endoscopic examinations are considered the gold standard for evaluating swallowing disorders; however, they carry high risks and can be challenging to perform in patients with advanced ALS who have respiratory dysfunction and other conditions. Therefore, non-invasive and easy-to-observe assessment methods have been explored. Tongue pressure and tongue ultrasonography are representative methods; however, they are limited in their ability to adequately evaluate the pharyngeal phase, such as pharyngeal residue, while reflecting the oral phase, such as oral passage and oral residue, of swallowing (7, 20).

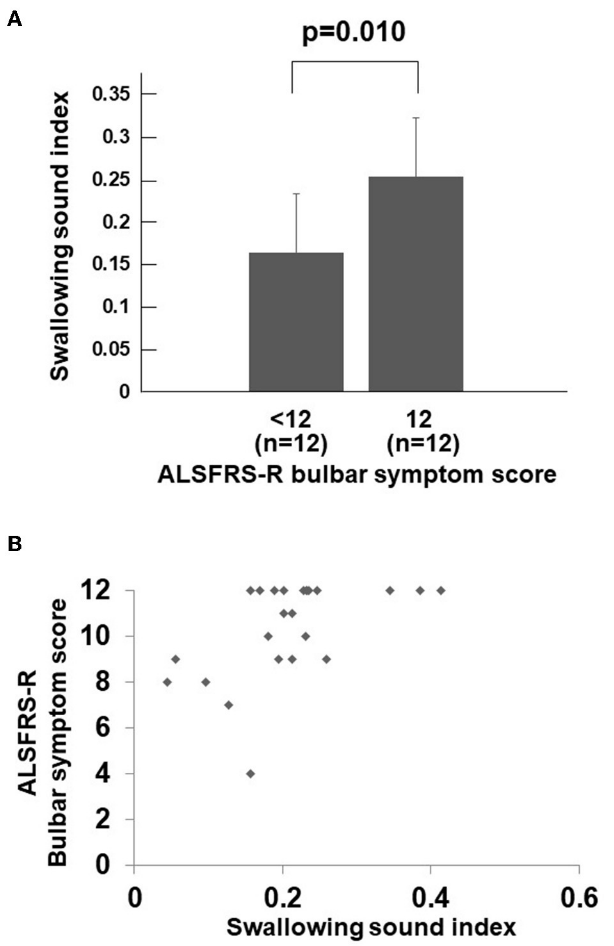

FIGURE 2
Swallowing sound index and ALSFRS-R bulbar symptom score. **(A)** Swallowing sound index between the ALSFRS-R bulbar symptom score <12 group and full score group. The swallowing sound index is significantly lower in the bulbar sub-score <12 group ($p = 0.010$). **(B)** Scatter plots of swallowing sound index and the ALSFRS-R bulbar symptom score. A positive correlation can be observed between the two, indicating a decrease in swallowing sound index with the bulbar symptoms. When the ALSFRS-R bulbar symptom score is less than 12 (full score), the swallowing sound index rapidly decreases. ALSFRS-R, Amyotrophic Lateral Sclerosis Functional Rating Scale-Revised.

TABLE 2 Factors associated with the swallowing sound index.

	Univariate analysis	Multivariate analysis		
Factors	*p*-value	coefficient	95% CI	*p*-value
Age	0.065			
Sex	0.388			
Body mass index	0.492			
Disease duration	0.052			
ALSFRS-R score	0.009*	0.006	0.003 – 0.010	0.002*
ALSFRS-R bulbar symptom score	0.007*	0.019	0.003 – 0.035	0.022*
Onset type	0.169			
Serum albumin	0.18			
% Vital capacity	0.002*	0.002	0.001 – 0.004	0.003*
Tongue pressure	0.004*	0.003	0.001 – 0.005	0.009*
MASA score	<0.001*	0.003	0.001 – 0.005	0.001*
MASA pharyngeal phase-related score	<0.001*	0.011	0.005 – 0.017	<0.001*

CI, confidence interval; ALSFRS-R, Amyotrophic Lateral Sclerosis Functional Rating Scale-Revised; MASA, Mann Assessment of Swallowing Ability. Multivariate analyses were performed using the individual factors, which showed a p-value of <0.05, in univariate analyses and age. *p-value of <0.05.

Although the origin and clinical significance of swallowing sounds are not yet clearly understood, they gradually disappear as swallowing disorders progress due to the decrease in swallowing pressure. Decreased swallowing sounds are suggestive of swallowing disorders. It has been suggested that the origin of swallowing sounds is the sound of the bolus passing through the open esophageal entrance; as such, they are a measure reflecting the pharyngeal phase, which is not contradictory (21, 22). The swallowing sound index calculated only the bolus inflow sounds among the various noises produced during the pharyngeal phase and was independently associated with the MASA pharyngeal phase score in this study. The MASA pharyngeal phase score reflects swallowing reflex, pharyngeal elevation, and pharyngeal clearance (18). Thus, the swallowing sound index may be an indicator of pharyngeal phase dysfunction. In this study, AI analysis revealed that a decreased swallowing sound index in patients with ALS was associated with a low bulbar symptom sub-score. Previously, it was reported that a low bulbar symptom sub-score in ALS patients reflected the delay of oral transit time and pharyngeal transit time using videofluoroscopic examination (20). These results further supported the notion that the swallowing sound index reflects the presence of dysphagia in ALS patients. In addition, the swallowing sound index had a correlation with tongue pressure, an existing evaluation method. Tongue pressure has been also reported as a sensitive marker for the early detection of swallowing disorders in spinal and bulbar muscular atrophy, which is also a motor neuron disease (23). ALS, a representative motor neuron disease, is characterized by a widespread loss of motor neurons throughout the body (12). Therefore, it is anticipated that swallowing difficulties are widely affected in both the oral and pharyngeal stages (20). Given this, it is important to develop an instrument to detect a disorder of the pharyngeal phase, such as by measuring the swallowing sound index using an electronic stethoscope. Moreover, it was also shown that the swallowing sound index tended to decrease along with %VC. The respiratory function primarily relies on the diaphragm and intercostal muscles, and it is reasonable to interpret that the correlation between the swallowing sound index and respiratory function is due to the widespread loss of motor neurons throughout the body, leading to a parallel decrease in both measures. However, in basic research, many of the swallowing-related muscles are derived from the branchial arches, and studies suggest that respiratory neurons are activated during swallowing (24). Therefore, it is possible that the decrease in the swallowing sound index could lead to a decrease in %VC. Based on these results, the AI analysis of swallowing sounds using an electronic stethoscope can be a simple method for assessing swallowing function, including the pharyngeal phase.

The usefulness of electronic stethoscopes and AI analyses during the COVID-19 pandemic is highly regarded. Using a stethoscope, patients can record their own body sounds and remotely evaluate them, thus AI analysis is extremely meaningful in terms of infection prevention measures. Furthermore, home care and monitoring are crucial for patients with debilitating diseases, such as ALS. Therefore, the evaluation method introduced in this study is considered very useful and significant.

This study had several limitations. First, it was conducted at a single facility. ALS is a rare disease, making it difficult to conduct studies with a large number of cases; therefore, further studies involving multiple facilities and more cases are needed in future. Second, there has been no comparative study yet that includes the gold-standard videofluoroscopic examination. In patients with ALS, the videofluoroscopic or endoscopic examination itself carries the risk of aspiration; therefore, a careful selection of cases is required for its implementation. However, it is necessary to establish the usefulness of these new testing methods. It is important to optimize the interval of analysis using videofluoroscopic examination to improve the accuracy of swallowing sound evaluation.

Swallowing sound evaluation using an electronic stethoscope AI analysis showed a correlation with existing indicators in swallowing evaluation in patients with ALS and suggested its usefulness as a new method. It is expected to be useful for patients with neurological disorders and the elderly who are at risk of aspiration. Furthermore, it is expected to be a useful examination method for home and remote medical care.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving human participants were reviewed and approved by the Ethics Committee of Hiroshima University Hospital. The patients/participants provided their written informed consent to participate in this study.

Author contributions

MN, RI, TW, MT, HN, and YS conceived and designed the study. MN, MT, TT, and YS performed the experiments. MN, TT, and YS analyzed the data. MN, RI, TW, MT, HN, and YS drafted the manuscript. YY and HM supervised the study. All authors contributed to the article and approved the submitted version.

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Conflict of interest

MN reported honoraria from Eisai, Takeda Pharmaceutical, Otsuka Pharmaceutical, Nihon Pharmaceutical, Sumitomo Pharma, Kyowa Kirin, Ono Pharmaceutical, AbbVie GK, and FP Pharmaceutical. HM reports honoraria from Eisai, Pfizer, Takeda Pharmaceutical, Otsuka Pharmaceutical, Nihon Pharmaceutical, Teijin Pharma, Sumitomo Pharma, Daiichi Sankyo, Kyowa Kirin, Novartis, Mitsubishi Tanabe Pharma, Ono Pharmaceutical, Biogen, and Bristol-Myers Squibb, AbbVie GK, Chugai Pharmaceutical, CSL Behring, UCB Japan, and research support from Eisai, Takeda Pharmaceutical, Otsuka Pharmaceutical, Nihon Pharmaceutical, Shionogi, Teijin Pharma, Fuji Film, Sumitomo Dainippon Pharma, Nihon Medi-Physics, Daiichi Sankyo, Kyowa Kirin, Sanofi, Novartis, Kowa Pharmaceutical, and Tsumura, Japan Blood Products Organization.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Findings of a videofluoroscopic swallowing study in patients with dysphagia

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Objective: Swallowing examination is crucial in patients with dysphagia. We aimed to compare qualitative and quantitative videofluoroscopic swallowing study (VFSS) results to provide reference for standardizing quantitative parameters.

Materials and methods: In total, 117 patients with dysphagia were included, 38 with Parkinson's disease and 39 and 40 in convalescence following cerebral hemorrhage and infarction. VFSS was both qualitatively and quantitatively analyzed.

Results: A significant difference of Oral transit time was found between the oral motor function grades ($p < 0.001$), also was swallowing reaction times found between swallowing reaction duration grades ($p < 0.001$), and soft palate lift duration between the soft palate lift grades ($p < 0.001$). Superior hyoid bone movement ($p < 0.001$), anterior hyoid bone movement ($p < 0.001$), hyoid pause time ($p < 0.001$), and hyoid movement duration ($p = 0.032$) had significant differences between the hyoid laryngeal complex movement grades, as did the pharyngeal cavity transit time among the cricopharyngeal muscle opening duration grades ($p < 0.001$). The laryngeal vestibule closure duration differed among the glottic closure grades ($p < 0.001$). No statistically significant difference in upper esophageal sphincter opening diameter ($p = 0.682$) or duration ($p = 0.682$) among the cyclopharyngeal muscle opening duration grades. The pharyngeal area at rest did not significantly differ among the different vallecular residue ($p = 0.202$) and pyriform sinus residue ($p = 0.116$) grades.

Conclusion: Several quantitative parameters can reflect the swallowing assessment process well. Further optimization of quantitative parameters is recommended.

KEYWORDS

dysphagia, videofluoroscopic swallowing study, qualitative analysis, quantitative analysis, parameters

1. Introduction

Dysphagia is a term that describes abnormal swallowing function caused by various factors in different parts of the body, with a prevalence ranging from 19% ~ 81% (1). The incidence of dysphagia caused by stroke in the acute phase was shown to be approximately 46.3%; during the period of convalescence, however, the incidence increased to 56.9 ~ 81.0% (2). The incidence rate of dysphagia varies in those with progressive neurological conditions. In patients with Parkinson's disease (PD), the incidence rate is approximately 50 ~ 87.1%; however, the risk of dysphagia in elderly patients with PD is about twice that in young patients, and the risk of dysphagia in patients with PD with a higher Hoehn and Yahr stage is about three times that in patients with a lower stage (3, 4). The main clinical manifestations of dysphagia include weakening of muscles involved in chewing, leading to food remaining in the mouth or leaking out of the mouth after swallowing, cough, reflux, and aspiration after swallowing (5). Related complications also include the consequences of aspiration (such as pneumonia, repeated cough, and asphyxia) and changes in diet and fluid intake (such as malnutrition, dehydration, decreased quality of life, and social isolation) (1). A literature review and analysis revealed that a series of complications caused by dysphagia irreversibly affected the physical and mental health of patients and their families to a certain extent (6). Ensuring that patients with dysphagia receive timely and correct examination and undergo appropriate and effective treatment and rehabilitation can promote functional recovery and reduce the occurrence of complications (7).

A videofluoroscopic swallowing study (VFSS) is a special type of examination involving X-ray fluoroscopy that is used to assess the movement of components involved in swallowing, including the mouth, pharynx, larynx, and esophagus. Analysis of VFSS data can lead to the discovery of swallowing function abnormalities from spot film and video, including further frame-by-frame and slow playback analysis. This examination is a widely used and relatively mature technique for assessing the swallowing function of patients in clinical practice, as it can directly reflect dynamic changes in the functions of the participating organs. VFSS is mainly used to assist in the diagnosis of swallowing disorders and is considered to be the "ideal method" for swallowing disorders examination, and it remains the "gold standard" for diagnosis (8).

The qualitative analysis of VFSS data mainly involves the assessment of the presence of aspiration and the evaluation of tongue movement, cricopharyngeal muscle function, swallowing reflex, laryngeal lift, epiglottic vallecula, and/or pyriform fossa retention via angiography to determine the swallowing function of patients (9, 10). Currently, qualitative analysis is the most widely used method for clinical evaluation, owing to its simplicity and high efficiency. However, there are certain shortcomings related to qualitative analyses. For example, the comprehensiveness and accuracy of the evaluation content are dependent on the patient's cooperation during and the methodology of the imaging examination, the quality of the recorded video, the technical expertise of the imaging personnel, and the analytical aptitude and experience of the rehabilitation physician or therapist evaluating the findings. Therefore, qualitative assessments fail to satisfy the criteria required for objective clinical evaluations and scientific research.

With the progressive advancements in imaging technology, a dynamic swallowing method has been developed to analyze the VFSS in recent 30 years (11, 12). The scholars have attempted to conduct quantitative VFSS analyses by recording the imaging data using a digital acquisition system at a speed of 30 frames/s, browsing it frame-by-frame, and quantifying the temporal and kinematic parameters involved in the swallowing process (13, 14). Further standardization and validation is required before such methods can be widely used in quantitative clinical evaluations and scientific research involving patients with dysphagia. Therefore, this study aimed to conduct a comparative analysis of quantitative and qualitative VFSS findings among patients with clinical dysphagia to obtain more valuable information and then use analysis software to quantify and automatically analyze the swallowing function of patients and apply it to clinical practice.

2. Materials and methods

2.1. Study design and participants

The participants were selected from patients with dysphagia who were treated in the Speech Rehabilitation Department of Beijing Rehabilitation Hospital affiliated with Capital Medical University from June 2022 to December 2022. The inclusion criteria were as follows: (1) patients who experienced cerebral hemorrhage and cerebral infarction who met the relevant diagnostic criteria formulated by the Fourth National Conference on Cerebrovascular Diseases (1995) and received a definitive diagnosis of stroke based on head magnetic resonance imaging (MRI); (2) patients with PD who met the MDS clinical diagnostic criteria for Parkinson's disease (15); (3) patients with varying degrees of comorbid dysphagia based on Expert consensus on evaluation and treatment of swallowing disorders in China (2017); (4) those with stable vital signs who were able to pay good attention to their surroundings, exhibited no serious cognitive impairment, and were able to cooperate to complete the required angiography. The exclusion criteria were as follows: (1) patients with complete or severe dysphagia; (2) those with an allergy to the contrast agent; (3) those with dysfunction of the heart, kidney and other organs; (4) patients with organic lesions of the esophagus, pharynx, and mouth; (5) those with a recent history of treatment with muscle relaxants and sedatives that could affect swallowing function; (6) individuals with cognitive impairment and mental illness; and (7) those with thyroid disease, local infection, other local diseases of the throat, or chronic respiratory diseases.

2.2. Sample size

We calculated the required sample size based on previous research of the incidence of stroke in the acute phase, stroke in convalescence and Parkinson's disease. Based on an alpha level of 0.05 and a power of 0.90, we estimated that we would require a total of 60 subjects, divided among three groups.

2.3. Videofluoroscopic swallowing study methodology

All patients underwent VFSS examination with an OPERA digital multifunctional gastrointestinal imaging device (GMM Group). Images were jointly collected by an radiologist with many years of diagnostic experience and an experienced rehabilitation physician with experience treating dysphagia. Each patient swallowed a 5 mL volume and medium consistency of food paste, containing a thickener and an iodohydrin contrast agent. We believe that this choice is appropriate in terms of safety, with a moderate degree of risk of aspiration, meanwhile it can also cause a series of swallowing movements, the quantitative data can be collected well, which basically meets the needs of this study. The state of the food was recorded as it passed through the mouth, pharynx, and esophagus in the anteroposterior and lateral orientations through video fluoroscopy. In VFSS, the swallow was performed once.

2.4. Qualitative analysis methods

According to the VFSS, abnormal swallowing function was assessed and the related physiological and pathological components were analyzed, including the oral motor function, swallowing reaction function, soft palate lift function, hyoid–laryngeal complex movement, cricopharyngeal muscle opening duration, glottic closure, and the presence of vallecular and pyriform sinus residues. A double-blind evaluation of the VFSS findings was conducted by a PhD candidate studying speech rehabilitation and a professional physician with many years of experience in diagnosing dysphagia. The two doctors independently completed the evaluation. Any discrepancies between the two researchers were reconciled by discussion between these two individuals before determining the final results. Eight qualitative components related to swallowing function were graded (16) and scored as listed in Table 1.

TABLE 1 Grades and scoring of the qualitative items.

Item	Grade	Score
Oral motor	Normal	3
	Impaired	2
	Severe impaired	1
Swallowing reaction	Normal	2
	Delayed	1
Soft palate lift function	Normal	3
	Impaired	2
	Severe impaired	1
Hyoid laryngeal complex movement	Intact	3
	Inadequate	2
	None	1
Cricopharyngeal muscle opening duration	Normal	3
	Delayed	2
	Severe delayed	1
Glottic closure	Intact	3
	Inadequate	2
	None	1
	Unable to cooperate	0
Vallecular residue	None	3
	<50%	2
	> 50%	1
Pyriform sinus residue	None	3
	<50%	2
	>50%	1

2.5. Quantitative analysis methods

The temporal and kinematic parameters of the VFSS were collected and recorded in a double-blind manner by a speech rehabilitation physician in the field of dysphagia and a speech rehabilitation therapist with extensive experience in the treatment of dysphagia. The average of each value determined by the two evaluators was used as the final result. If the two values differed, they were discussed, and the final result was determined through negotiation.

An 8 mm reference ball was set up as calibration index for calculating distances in the analysis of kinematic parameters. Quantitative VFSS analysis contains four kinematic parameters and eight temporal parameters. The specific kinematic parameters were assessed including the following: (1) hyoid bone superior movement (HSM), the vertical distance between the lowest and highest positions of the hyoid bone during the swallowing process, from the movement of the hyoid bone to its return to its original position and (2) hyoid bone anterior movement (HAM), the horizontal distance between the lowest and highest positions of the hyoid bone (17, 18). The interception method is shown in Figures 1A,B. The line between the lower anterior corners of C2 and C4 represented the vertical axis, which was made neutral by rotation and lied perpendicular to the horizontal axis of the image, C2 and C4 vertebrae shown in Figures 1A,B, then sent their coordinates (x1, y1), (x2, y2), (C4x1, C4y1), (C4x2, C4y2) to the analysis software, and the hyoid bone movement was calculated using the built-in Equations (1) and (2).

$$\text{HAM} = (x2 — x1) — (\text{C4x2} — \text{C4x1}), \tag{1}$$

$$\text{HSM} = (y2 — y1) — (\text{C4y2} — \text{C4y1}), \tag{2}$$

The kinematic parameters also included: (3) the upper esophageal sphincter (UES) opening diameter, as shown in Figure 2, the width of the narrowest part of the pharyngoesophageal sphincter in the lateral image at the maximum degree of expansion induced by the mass in a single swallow, with the measurement line perpendicular to the edge of the spine (19); and (4) the pharyngeal area at rest, the minimum lateral area of the swallowed mass and pharyngeal cavity upon contraction in one mouthful (20).

The eight time parameters assessed have been included as follows: (1) the oral transit time, defined as the time interval between the food completely entering the mouth and being pushed by the tongue muscle resulting in a change in shape until the time at which the head of the food ball reached the intersection between the mandibular branch and the base of the tongue (21); (2) the soft palate elevation time, the interval between the time when initial soft palate contact and the time at which the posterior pharyngeal wall moves down to its original position (22); (3) the hyoid at rest duration, the length of time when the hyoid bone is completely at rest position during swallowing (22); (4) the hyoid movement duration, defined as the time interval between the initiation of the forward and downward movement of the hyoid bone and the return of the hyoid bone to its resting position (18); (5) the UES opening duration, representing the time interval between the beginning of

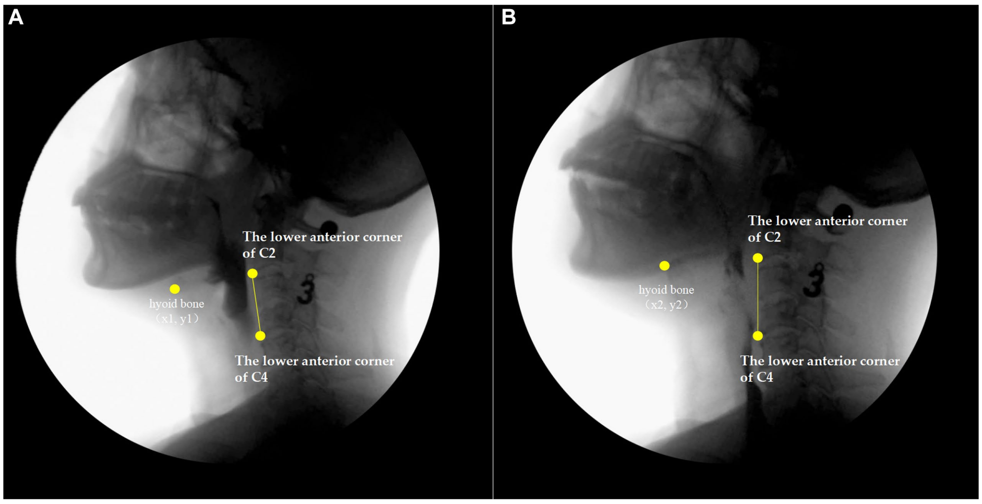

FIGURE 1
Hyoid bone superior and anterior movement based on videofluroscopy. **(A)** screenshot showing the initial state of the hyoid bone after rotation; **(B)** screenshot of the time point at which the hyoid bone has been lifted to the highest and farthest position after rotation. x1 is the horizontal coordinate of the hyoid resting position, and y1 is the vertical coordinate of the hyoid resting position; x2 is the abscissa of the farthest point of hyoid motion, and y2 is the ordinate of the farthest point of hyoid motion; C4xl is the abscissa of the lower anterior corner of C4 in the hyoid resting position, and C4yl is the ordinate of the lower anterior corner of C4 in the hyoid resting position; C4x2 is the abscissa of the lower anterior corner of C4, the farthest point of hyoid motion, and C4y2 is the ordinate of the lower anterior corner of C4, the farthest point of hyoid motion.

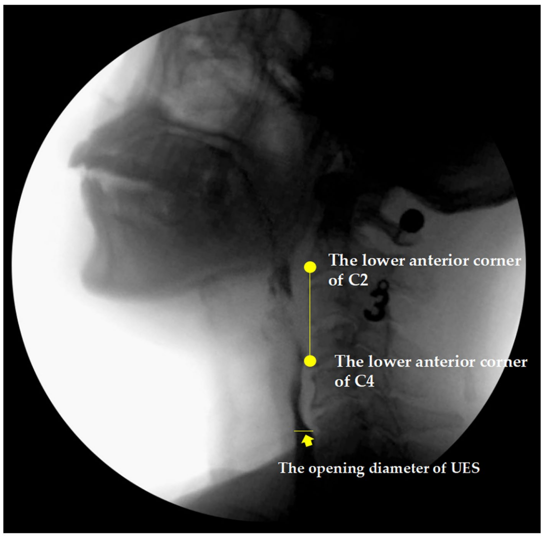

FIGURE 2
The upper esophageal sphincter (UES) based on videofluroscopy.

the opening and the complete closing after the mass reaches the UES (14); (6) the swallow reaction time, which is the time interval between the head of the mass reaching the intersection of the lingual and mandibular branches and the start of the swallowing phase, marked by the initiation of hyoid bone movement (14); (7) the pharyngeal transit time, defined as the time interval between the head of the food mass passing the intersection of the lingual and mandibular branches and the tail of the food mass passing the UES (23); and (8) the laryngeal vestibule closure (LVC) duration, representing the time interval between the closing and reopening of the laryngeal vestibule (23).

2.6. Statistical analysis

Statistical Package for the Social Sciences (SPSS) version 22.0 software was used for statistical analysis. One-way ANOVA and Bonferroni post-hoc tests were used to assess the differences in the mean age or the mean score of the clinical groups. Kruskal–Wallis test was used for comparison among multiple groups. $p < 0.05$ is statistically significant. The measurement data conforming to a normal distribution is expressed as mean ± standard deviation, and the measurement data not conforming to a normal distribution is expressed as median (25th percentile–75th percentile).

To evaluate different dimensions of the qualitative values, principal component analysis (PCA) was performed. PCA is concerned with establishing which linear components exist within the data and how a particular variable might contribute to that component. The PCA was conducted with all 12 items into analysis to maximize the loadings of the variables onto one factor (the factor that intersects the cluster) and minimize them on the remaining factor(s). The assumptions were fulfilled (Bartlett's test was highly significant and the Kaiser–Meyer–Olkin (KMO) measure of sampling adequacy > 0.6). The analysis of the association between the qualitative results and quantitative results was performed using Pearson's correlation coefficients.

3. Results

3.1. Overview

A total of 151 patients with dysphagia were considered for inclusion, 123 of whom agreed to participate. Of these 123 patients, three did not meet the inclusion criteria (one case had a respiratory infection and two cases experienced a recurrence of sudden cerebral

TABLE 2 The demographic characteristics of the patients with dysphagia.

Variables	Patients
Sample size	117
Sex (male/female)	61/56
Mean age (years)	61.31 ± 12.38 (27–83)
Type	
Parkinson's disease	38
Cerebral hemorrhage	39
Cerebral infarction	40

infarction) and three temporarily refused to undergo VFSS. Ultimately, 117 patients with dysphagia completed the study and were included in the analysis. Table 2 summarizes the detailed demographic data of the participants and the cause of dysphagia.

3.2. Qualitative grading and quantitative value matching of VFSS

The mean and standard deviation values of the quantitative items according to the different grades of the qualitative items of the VFSS are listed in Table 3. Among these items, the oral transit time significant differed among the different grades of qualitative oral motor function; with poorer oral motor function resulting in significant prolongation of the oral transit time. The quantitative swallowing reaction time significantly differed among the qualitative swallowing reaction grades; that is, the worse the swallowing reaction grade, the longer the swallowing reaction time. Similarly, as the grade of the soft palate lift decreased, the soft palate lift duration was significantly prolonged. Weakening of the hyoid–laryngeal complex movement resulted in significant shortening of the HSM and HAM and prolongation of the hyoid pause time and hyoid movement duration. Poorer cricopharyngeal muscle opening durations resulted in significant prolongation of the pharyngeal cavity transit time, poorer glottis closure grades resulted in significant prolongation of the LVC duration.

The UES opening diameter and duration both decreased as the grade of the cricopharyngeal muscle opening duration increased, although neither parameter significantly differed between the different qualitative grades of cricopharyngeal muscle opening. There was no statistically significant difference in the pharyngeal cavity contraction rate among the different qualitative grades of the presence of vallecular and pyriform sinus residues. Almost all patients with dysphagia exhibited vallecular residues (96.58%); of them, 53.10% had a small amount, whereas 46.90% had a large amount. No vallecular residue was observed in 4.27% of participants. Approximately 4/5 patients with dysphagia exhibited pyriform sinus residues (83.76%); of them, 48.98% had a small amount and 51.02% had a large amount.

3.3. The qualitative total value of VFSS corresponds to the quantitative value

The mean and standard deviation values of the quantitative kinematic parameters for the different qualitative total value categories are listed in Table 4. Among them, the HSM, HAM, and pharyngeal area at rest significantly differed according to the different qualitative total value classifications. However, the opening diameter of the UES did not significantly differ between the qualitative total value classifications, although the range decreased as the qualitative total value classification worsened.

For the time parameters, the mean and standard deviation values of the quantitative measures for each qualitative total value range are listed in Table 5. Every quantitative measure of the time parameters significantly differed according to the qualitative total value classification. As the qualitative total grading worsened, the oral transit time, soft palate lift duration, hyoid pause time, hyoid movement duration, swallowing reaction time, pharyngeal cavity transit time, and LVC duration were significantly prolonged. The opening duration of the UES was not significantly different between the groups with good and poor qualitative values; however, it was significantly longer than that in the group with poor results.

3.4. The correlation between the quantitative total value and qualitative total value

Bartlett's test of sphericity was highly significant ($p < 0.001$), indicating that correlations between the items were sufficiently high for PCA analysis. The KMO measure verified the sampling adequacy for the analysis, and KMO resulted in a value of 0.604, which is in agreement with the recommended assumptions. An initial analysis was run to obtain eigenvalues for each component in the data. Five components had eigenvalues over Kaiser's criterion of 1 and this combined explains 69% of the variance. Then the factor expression of quantitative total value is obtained, and the quantitative total value is calculated. The correlation between the qualitative total value and the quantitative total value was statistically significant ($p = 0.036 < 0.05$), the correlation coefficient was 0.194, which shows that there is no relevance between them.

4. Discussion

The swallowing process involves a series of complex, highly coordinated, and fixed muscle movement behaviors. Applying instrument to evaluate swallowing can more directly and accurately evaluate the swallowing situation in oral, pharyngeal and esophageal stages, and understand the integrity of the protection function of the swallowing airway. It is significance for the diagnosis, selection of intervention methods, and management of Dysphagia in the swallowing stage. VFSS and flexible endoscopic examination of swallowing (FEES) are the gold standard to determine Dysphagia. In addition, pharyngeal cavity pressure measurement is widely carried out in recent years, which can measure the pressure in the pharyngeal cavity and quantify the swallowing function, such as high-resolution pharyngeal cavity pressure measurement (HRM), upper esophageal Sphincter pressure measurement, and automatic pharyngeal impedance pressure measurement.

Compared to VFSS, the above evaluation methods have their unique advantages and unavoidable drawbacks. FEES can observe the transport of food pellets to the throat during swallowing under direct

TABLE 3 Qualitative grading and quantitative value matching of videofluoroscopic swallowing study (VFSS).

Qualitative items	Grade	*n*	Quantitative items	*F*	*p*
			Oral transit time (s)		
Oral motor	Normal	59	2.75 ± 1.95	52.103	<0.001
	Impaired	34	5.40 ± 3.41		
	Severe impaired	24	11.46 ± 5.94		
			Swallowing reaction time (s)		
Swallow reaction	Normal	44	1.00 ± 0.99	4.207	<0.001
	Delayed	72	0.55 ± 0.54		
			Soft palate elevation time (s)		
Soft palate lift	Normal	81	1.99 ± 1.17	34.508	<0.001
	Impaired	20	2.17 ± 1.43		
	Severe impaired	16	8.24 ± 6.99		
			HSM (mm)		
Hyoid laryngeal complex movement	Intact	24	10.55 ± 7.60	9.058	<0.001
	Inadequate	51	7.97 ± 6.52		
	None	42	4.41 ± 3.45		
			HAM (mm)		
	Intact	24	9.28 ± 5.59	9.801	<0.001
	Inadequate	51	14.45 ± 7.10		
	None	42	16.07 ± 4.88		
			Hyoid at rest time (s)		
	Intact	24	0.11 ± 0.07	7.479	0.001
	Inadequate	51	0.08 ± 0.06		
	None	42	0.06 ± 0.03		
			Hyoid movement duration (s)		
	Intact	24	1.72 ± 0.50	3.551	0.032
	Inadequate	51	2.14 ± 1.11		
	None	42	2.59 ± 1.76		
			UES opening diameter (mm)		
Cricopharyngeal muscle opening duration	Normal	58	6.93 ± 2.34	0.384	0.682
	Delayed	35	6.67 ± 1.56		
	Severe delayed	24	6.56 ± 1.85		
			UES opening duration (s)		
	Normal	58	0.90 ± 0.25	1.823	0.166
	Delayed	35	0.90 ± 0.34		
	Severe delayed	24	0.78 ± 0.27		

(Continued)

TABLE 3 (Continued)

Qualitative items	Grade	*n*	Quantitative items	*F*	*p*
			Pharyngeal cavity transit time (s)		
	Normal	58	1.54 ± 0.67	15.540	<0.001
	Delayed	35	1.64 ± 0.86		
	Severe delayed	24	2.89 ± 1.68		
			LVC duration (s)		
Glottic closure	Intact	68	0.71 ± 0.38	8.343	<0.001
	Inadequate	22	1.07 ± 0.89		
	None	12	1.44 ± 1.05		
	Unable to cooperate	15	/		
			Pharyngeal area at rest (%)		
Vallecular residue	None	5	36.60 ± 17.39	1.619	0.202
	<50%	60	44.13 ± 30.16		
	>50%	53	36.38 ± 12.19		
			Pharyngeal area at rest (%)		
Pyriform sinus residue	None	19	41.75 ± 15.39	0.116	0.890
	<50%	48	41.44 ± 27.14		
	> 50%	50	38.90 ± 18.97		

VFSS: video fluoroscopic swallowing study; HSM: hyoid bone superior movement; HAM: hyoid bone anterior movement; UES: upper esophageal sphincter; LVC: laryngeal vestibule closure.

view of the monitor, and observe the deformation and displacement of food pellets in the throat. Therefore, FEES can better reflect the anatomical structure of the throat and the accumulation of food masses compared to VFSS, and is more suitable for swallowing dysfunction caused by cranial neuropathy, postoperative or traumatic injuries, and anatomical structural abnormalities. It is also suitable for research on aspiration (24, 25). Another advantage of FEES is that it has no X-ray radiation and can be repeatedly checked. The device is easy to carry and can be checked by the bedside. However, FEES cannot directly observe the entire process of food mass transportation, and can only judge the swallowing effect through indirect information on the distribution of food mass in the pharynx after swallowing. Without hunger, the opening of the cricopharyngeal muscle can be directly observed. Therefore, it cannot directly evaluate the coordination between swallowing organs. When the swallowing amount reaches its maximum or the food covers the laryngoscope lens, it will not be imaged. HRM can dynamically and continuously reflect the changes in pharyngeal pressure throughout the swallowing process, with a focus on reflecting pharyngeal coordination. The disadvantage is that it is not possible to directly see the anatomical structure and food passage status, nor can it determine whether there is aspiration (26). Meanwhile, VFSS still has certain drawbacks. If it wants to receive X-ray radiation, it needs to be transferred to the radiology department, which cannot reflect the sensory function of the pharynx. The most important thing is that it cannot quantitatively analyze the pharyngeal

TABLE 4 Comparison to quantitative values of motion parameters (mm) by qualitative total value.

Qualitative total value	*n*	HSM	HAM	UES opening diameter	Pharyngeal area at rest (%)
Good (24′–20′)	29	4.83 ± 3.98	10.17 ± 6.33	7.20 ± 1.38	41.80 ± 18.51
Passable (19′–13′)	68	8.50 ± 7.74	14.83 ± 6.59	6.70 ± 1.90	41.35 ± 24.74
Poor (12′–8′)	20	8.87 ± 3.65	16.31 ± 4.52	6.20 ± 1.04	55.00 ± 9.54
F		3.745	7.438	1.225	3.304
p		0.027	<0.001	0.230	0.040

HSM: hyoid bone superior movement; HAM: hyoid bone anterior movement; UES: upper esophageal sphincter.

muscle contraction force and the pressure inside the food mass, and much of the information recorded during VFSS cannot be fully utilized. However, the location can be determined and symptoms of dysphagia can be observed by angiography during swallowing. Previous studies have confirmed that dynamic contrast quantitative analysis technology can effectively clarify the relationship between the movements of the organs involved in swallowing as a food bolus passes (27–29). Applying VFSS to the analysis of muscle relaxation and contraction of the upper sphincter of the esophagus and pharynx can provide more detailed information than can be determined by assessments based on contrast alone. Determining the relationship between quantitative and descriptive findings could be useful in clinical practice, although it was previously unknown whether such outcomes would consistently match up. Therefore, the VFSS qualitative and quantitative results were compared and analyzed in this study.

The quantitative values of the kinematic parameters differed based on the qualitative grades of the VFSS. First, weakening of the movement of the hyoid–laryngeal complex resulted in lower HSM and HAM values, confirming the weaker the movement of the hyoid laryngeal bone complex, the greater the degree of swallowing dysfunction. This finding is consistent with the results of other studies that have investigated swallowing physiology and pathology (17, 30). Movement of the hyoid–laryngeal complex is a crucial component of swallowing function, as it helps ensure the closure of the throat, the return of the epiglottis, the opening of the cricopharyngeal muscle, and the smooth and safe completion of swallowing activities. Measuring the displacement of the hyoid bone is often used to quantify the movement ability of the hyoid–laryngeal complex. During swallowing, the vertical movement of the hyoid bone drives the closure of the epiglottis, which is beneficial for the protection of the airway, whereas the forward movement of the hyoid is beneficial for the opening of the UES (31). Theoretically, upward and forward displacement of the hyoid bone plays a positive role in swallowing. The results of this study also confirmed that the measuring the displacement of the hyoid bone can help to objectively evaluate the motion amplitude of the hyoid-laryngeal complex and can compensate for the lack of information provided in clinical evaluations based solely on observation of tongue extension or swallowing angiography to describe the motion of the hyoid-laryngeal complex.

An increase in the cricopharyngeal muscle opening duration, however, did not significantly alter the opening diameter of the UES. Similarly, the opening duration of the UES did not change significantly with the different qualitative total value categories. Physiologically, the coordination of activities involved in swallowing mainly the coordination of UES relaxation and pharyngeal muscle contraction, as well as the sequential movement of upper and lower pharyngeal muscle contractions. The evaluation of swallowing coordination via VFSS is mainly based on the observation and description of the opening of the cricopharyngeal muscle; however, such observations may be subjective, and no unified diagnostic criteria have been established. In contrast, quantitative analysis allows for the assessment of the opening range of the UES, which is an important quantitative index that reflects the coordination of the swallowing process (32). The quantitative analysis of the VFSS data facilitated the measurement of the opening range of the UES, although no obvious difference with qualitative total value; one possible explanation for this could be related to the primary disease associated with the dysphagia among the patients included in this study. Previous studies have reported that coordination of movements involved in swallowing is regulated by the swallowing pattern generator within the brainstem (33, 34). The development of a brainstem lesion usually manifests as weakening of the pharynx's ability to push and/or an abnormal UES relaxation function, which can easily lead to serious consequences that include leakage or aspiration. However, the present study did not include patients with such brainstem diseases; therefore, the value of the UES opening diameter does not reliably reflect the relaxation of the cricopharyngeal muscle.

There was no significant difference in the pharyngeal area at rest between the different qualitative grades of vallecular and pyriform fossa residues. The pharyngeal area at rest can be used as another objective index for evaluating the coordination of the pharyngeal phase of swallowing. The pharyngeal cavity contraction rate reflects the degree of contraction during swallowing in the pharyngeal phase (35, 36). In this stage, the hyoid bone on the larynx moves upward, the arytenoepiglottis and thyrohyoid muscles contract, and the base of the tongue inclines backward to ensure the epiglottis forms a proper cover; while the epiglottis valley on both sides is oriented close to the midline, the muscle group in the larynx contracts, the vocal cord and the ventricular band retracts, the glottis closes, and the pharyngeal constrictor retracts. During this time, the laryngopharynx and pyriform fossa are open, and the food mass is squeezed across the epiglottis, reaching the esophageal entrance; the opening of the upper esophageal sphincter is coordinated to ensure smooth passage of food through the pharyngeal cavity for entry into the esophagus. Thus, the main function of the pharyngeal cavity and the related muscle contraction is to clear the pharyngeal mass and squeeze the food bolus downward into the esophagus during swallowing. When swallowing disorders are caused by various organic and neuromuscular abnormalities, the ability of the pharyngeal cavity to clear food decreases, the corresponding size of the food mass remaining in the pharyngeal cavity increases, and the corresponding pharyngeal cavity contraction rate decreases. Aspiration can easily occur when the

TABLE 5 Comparison of quantitative values of time parameters by qualitative total value.

Qualitative total value	*n*	Oral transit time (s)	Soft palate lift duration (s)	Hyoid at rest duration (s)	Hyoid movement duration (s)	UES opening duration (s)	Swallow reaction time (s)	Pharyngeal cavity transit time (s)	LVC duration (s)
Good (24′–20′)	29	3.20 ± 3.14	1.72 ± 1.42	0.08 ± 0.06	1.72 ± 0.55	0.91 ± 0.31	0.52 ± 0.43	1.44 ± 0.51	0.59 ± 0.11
Passable (19′–13′)	68	3.93 ± 3.34	2.52 ± 2.20	0.08 ± 0.06	2.14 ± 1.11	0.92 ± 0.29	0.65 ± 0.57	2.01 ± 1.00	0.92 ± 0.78
Poor (12′–8′)	20	11.3 ± 10.0	8.98 ± 8.60	0.05 ± 0.02	3.20 ± 2.16	0.70 ± 0.17	2.24 ± 2.06	2.75 ± 2.22	1.02 ± 0.36
F		18.956	24.181	3.696	8.493	5.230	23.272	6.880	3.645
p		<0.001	<0.001	0.028	<0.001	0.007	<0.001	0.002	0.029

UES: upper esophageal sphincter; LVC: laryngeal vestibule closure.
No., number.

glottis reopens. Previous studies have shown that in the treatment of dysphagia, improving pharyngeal contraction can effectively reduce the residue remaining after swallowing (37). However, the pharyngeal cavity contraction rate in this study did not significantly correlate with the grading of epiglottic valley and pyriform fossa residues. Considering the limited inclusion of primary diseases in this study, measurements of the pharyngeal cavity area and contraction rate may have had little correlation with the presence of such residues. In addition, the results of this study revealed that 96.58% of the patients had vallecular residues, 83.76% had pyriform fossa residues, and the average contraction rate of the pharyngeal cavity was 40–55%. Therefore, it is also possible that the 5 mL volume of food paste administered in this study was too small, which could have resulted in weak sensory and motor stimulation of the pharynx, thereby affecting the contraction of the pharyngeal constrictor muscle and resulting in insufficient peristalsis.

In this study, most of the quantitative and time parameter values showed statistically significant differences according to the different qualitative grades assigned during the VFSS, including the oral transit time, swallowing reaction time in the pharyngeal phase, soft palate lift duration, hyoid movement duration, pharyngeal cavity transit time, and LVC duration. However, the UES opening duration poorly reflected the degree of cricopharyngeal muscle opening, which is consistent with the UES opening diameter results.

All quantitative values were measured or calculated built-in software tools and formulas, reflecting a portion of the time sequence and interval during the swallowing process. A factor analysis for dimensionality reduction of the 12 quantitative variables was conducted, and the results suggested that the quantitative items were relatively independent. The five principal components selected barely represented all of the quantitative values; that is, the measurements of the 12 quantitative variables could still adequately describe the entire swallowing process. The results of this study also show that there is a low correlation between the quantitative total value and the qualitative total value, which means quantitative results can not reveal that the correlation and sensitivity with qualitative results. However, since the types and definitions of the parameters used by various institutions are not yet unified, this study suggests that in the future selection of quantitative parameters of VFSS, studies should continue to optimize the existing parameters and attempt to screen out and standardize effective and comprehensive parameters to fully describe the swallowing process. This could help promote their use in clinical settings to better evaluate the effects on patients before and after treatment or the differences between patients.

The quantitative of VFSS based on pathological samples collected from a wide range of individuals with multiple diseases, the extraction of the most effective and valuable information from dysphagia angiography and the objective comparison of the levels of functionality within-patients before and after treatment, as well as between patients, can fully meet the comprehensive needs of scientific research, stimulate more innovative research, and generate ideas and references for the evaluation and follow-up treatment of dysphagia. For now, the quantitative analysis of VFSS is mainly used to describe the physiological state of swallowing (14, 38), explore the pathological and physiological characteristics of swallowing in different diseases (23, 39), analyze the effects of age, gender, texture of food balls and other factors on swallowing (40, 41), and evaluate treatment efficacy (42, 43). In the future, the quantitative results should be used for evaluation, and future studies should assess other valuable parameters and improve those with poor reliability, validity, and matching. With progress in science and technology and further deepening of research in this field, fully automated quantitative analysis of VFSS data could become possible, improving the effectiveness of swallowing assessments and reducing the burden on clinical workers.

5. Strengths and limitations

This study has the following limitations: (1) the types of patients with dysphagia selected in the study was relatively limited, the patients with PD were in phase 1–2, and the existing dysphagia was relatively mild, and the representativeness of the sample was relatively weak, so further research must be conducted in the future with an improved design; (2) the type of food balls selected was relatively fixed, which could have had a certain impact on the results; (3) the sample size needs to be further expanded; (4) during video acquisition, due to the patients' conditions, the body position, head control, and degree of cooperation could have been impacted, among other factors, which could have affected the clarity of imaging of various anatomical components, resulting in difficulties and errors in the qualitative analysis; (5) when obtaining various quantitative results, semi-automatic methodologies may lead to some measurement errors due to deviations of the measurer's understanding of the measurement technique, and the workload of the data acquisition process is large with many steps, so fatigue could have led to some measurement errors; and (6) considering the imaging factors, the radiation amplification effect could give the impression that the distance between two points on the image is larger than the actual value, and the radial distortion of the ray could stretch the length of the structure around the image. Radiation amplification and radial distortion of the rays may have affected the accuracy of the analysis.

6. Conclusion

In conclusion, there was a good match between the qualitative and quantitative VFSS time parameter values. However, the kinematic parameters did not accurately reflect the quantitative results. Determining quantitative values can still sufficiently describe the entire swallowing process, and these measures positively correlated with results of the qualitative evaluations. It is recommended that the quantitative evaluation parameters be optimized in future studies to facilitate assessments of swallowing function in patients with dysphagia.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding authors.

Ethics statement

The study was conducted in accordance with the Declaration of Helsinki and was approved by the Ethics Committee of Beijing Rehabilitation Hospital (protocol code 2020-008 and approved on December 1, 2020). Informed consent was obtained from all participants involved in the study. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

Author contributions

XD designed this study. QC, BW, WJ, and HW collected the data. QC analyzed the data and drafted the manuscript. YH and QZ interpreted and critically assessed the results. JX and XD further revised the manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Detailed findings of videofluoroscopic examination among patients with Parkinson's disease on the effect of cervical percutaneous interferential current stimulation

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Introduction: Parkinson's disease (PD) leads to various types of swallowing disorders. We investigated the effect of cervical percutaneous interferential current stimulation on dysphagia. By conducting detailed qualitative and quantitative analysis of videofluoroscopic examination, we aimed to understand dysphagia in patients with PD and investigate its effects on swallowing function.

Methods: Patients received cervical percutaneous interferential current stimulation for 20 min twice a week for 8 weeks. In this exploratory study, we evaluated aspiration/laryngeal penetration, oral cavity residue, vallecular residue, and pharyngeal residue. In addition, we performed temporal analysis.

Results: Twenty-five patients were completely evaluated. At baseline, the proportions of laryngeal penetration/aspiration, oral cavity residue, epiglottic vallecula residue, and pharyngeal residue were 40.0, 88.0, 72.0, 60.0, and 16.0%, respectively. Conversely, pharyngeal transit time, laryngeal elevation delay time, pharyngeal delay time, and swallowing reflex delay were nearly within the normal ranges. Cervical percutaneous interferential current sensory stimulation improved only oral cavity residue at the end of the intervention, from 88.0 to 56.0%.

Discussion: Patients with PD demonstrated remarkably high frequencies of residues in the oral and pharyngeal regions. The usefulness of cervical interferential current stimulation was partially demonstrated for oral cavity residue. Considering that PD exhibits diverse symptoms, further accumulation of cases and knowledge is warranted.

Trial registration: jRCTs062220013.

KEYWORDS

Parkinson's disease, dysphagia, interferential current sensory stimulation, videofluoroscopic examination, temporal analysis

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1. Introduction

Neurological disorders often accompany dysphagia, and dysphagia in patients with Parkinson's disease (PD) holds significant importance. Aspiration pneumonia, arising from dysphagia, represents a major cause of mortality among patients with PD, highlighting the unmet medical need in managing swallowing difficulties in this population. PD leads to the following types of swallowing disorders: abnormal transport from the oral cavity and pharynx (1), delayed swallowing reflex (2), pharynx residue (3), and others. In addition, silent aspiration, which is caused by decreased sensation in the pharynx and larynx, is also serious and characteristic in patients with PD (3).

We planned an intervention trial that primarily focused on silent aspiration, aiming to activate the sensory nerves through cervical percutaneous interferential current stimulation. Recent innovations in neurological stimulation methods include cervical percutaneous electrical stimulation to enhance neuromuscular function. Pulsed current approaches show promise in inducing muscle contractions to treat dysphagia but are associated with discomfort (4). An alternative method, interferential current sensory stimulation, activates peripheral nerves in the pharynx and larynx to heighten sensitivity and protect the airway (5). Reports suggest that electrical stimulation devices can enhance swallowing without muscle contraction (6, 7). Interferential currents penetrate deeper tissues comfortably compared to pulsed currents, holding potential for alleviating dysphagia (8). Studies have highlighted enhanced saliva production (9), reduced pharyngeal latency, increased swallowing frequency, and improved airway sensitivity (8). Cervical percutaneous interferential current stimulation might benefit patients with dysphagia by enhancing airway defense and nutrition (10). Moreover, cervical percutaneous interferential current can stimulate the central pattern generator (CPG) and improve the swallowing reflex (11). Previously, we investigated the relationship between videofluoroscopic examination (VF) and brain lesion sites in patients with stroke and reported a correlation between delayed swallowing reflex initiation and basal ganglia lesions (12). PD also involves abnormalities in the cerebral basal ganglia network. Therefore, the effectiveness of cervical percutaneous interferential current stimulation for the swallowing reflex might be considered.

In this study, the main objective was to assess the improvement of cough reflex in patients with PD. However, the complexity of dysphagia in PD arises from various factors, necessitating a comprehensive grasp of swallowing dynamics. We focused on exploring how cervical percutaneous interferential current stimulation impacts swallowing function in patients with PD. Through qualitative and quantitative analysis of VF, which is the gold standard method for evaluation, this study aimed to enhance the understanding of PD-related dysphagia and thoroughly examine the effects of cervical percutaneous interferential current stimulation.

Abbreviations: CPG, central pattern generator; LEDD, levodopa equivalent daily dose; LEDT, laryngeal elevation delay time; PD, Parkinson's disease; PDT, pharyngeal delay time; PTT, pharyngeal transit time; VF, videofluoroscopic examination.

2. Material and methods

2.1. Ethics approval, registration, and patient consent

This research received authorization from the Certified Review Board at Hiroshima University (<city>Hiroshima</city>, Japan) (approval ID: CRB6180006) and adhered to the directives of the federal administration in line with the principles outlined in the 1964 Declaration of Helsinki. It has been duly recorded in the jRCT database (jRCTs062220013). Comprehensive written consent was acquired from all participants involved in the study.

2.2. Study design and protocol

The study's design and protocol were previously published (13). The methodology consisted of a single-arm, open-label study that adhered to the reporting guidelines outlined in SPIRIT (14). Our investigation centered on assessing the effectiveness and safety of percutaneous neck interferential current stimulation in patients diagnosed with PD, as per the criteria set by the Movement Disorder Society, falling within Hoehn-Yahr stages 2–4 (15). The study was conducted at Hiroshima University Hospital.

We enrolled individuals who met the criteria of clinically probable or established PD as defined by the Movement Disorder Society criteria, with Hoehn-Yahr stages 2–4 at the time of registration. Additionally, participants needed to be capable of visiting the hospital twice weekly and provide informed written consent. Eligibility was restricted to patients aged between 19 and 86 years whose levodopa dosage had remained constant for over a month. Those with implanted pacemakers or defibrillators, undergoing deep brain stimulation, pregnant or attempting to conceive, diagnosed with or having a history of head or neck cancer, currently experiencing active pneumonia, or possessing a history of swallowing rehabilitation, were excluded from the study.

Participants underwent cervical interferential current stimulation for 20 min, twice a week, over an 8-week period. The stimulation was administered using a Gentle Stim® device from FoodCare Co., Ltd., Kanagawa, Japan. Electrode pads were applied to the front of the neck to stimulate the swallowing-related (glossopharyngeal nerve and superior laryngeal) nerves. A 50 Hz swallowing reflex interferential current stimulation was utilized because of its lower threshold in comparison to pulse stimulation, resulting in minimal sensation for patients. Stimulation adhered to a standardized protocol, with the maximum stimulation current set below the threshold at which the patient could perceive electrical sensations, ranging from 2.0 to 2.5 mA. Stimulation was administered consistently and repeatedly. Figure 1 shows the landscape of the stimuli.

Evaluations, except for VF, were conducted every 4 weeks from the start of the intervention to 16 weeks post-intervention initiation. VF assessments were conducted every 8 weeks within the same timeframe to minimize the radiation exposure.

The primary and secondary endpoints were outlined in a prior publication (13). As part of this study, we conducted a thorough and detailed evaluation using VF, a recognized gold

FIGURE 1
Picture of cervical percutaneous interferential current stimulation.

standard method for evaluating swallowing function. Our study concentrated specifically on VF findings.

2.3. Videofluoroscopic examination

An X-ray imaging system (Ultimax-i, CANON MEDICAL SYSTEM CORPORATION, Tochigi, Japan) was used and the tests were performed with patients in a seated position. The test material was 3 mL of water with 30%/w barium contrast medium (Barytester A240 Powder®, FUSHIMI Pharmaceutical Co. Ltd, Kagawa, Japan), which the patients were instructed to swallow after it was delivered via a syringe to the floor of the mouth. The evaluation using 3 mL of water is relatively widely accepted and implemented for assessing swallowing function (16). In addition, 3 mL should always be used in evaluations to prioritize safety in individuals with swallowing disorders (17). Another evaluation method, the simple water drinking test known as the Modified Water Swallow Test (18), also uses this volume of water. Therefore, the present study also used 3 mL of water. The imaging with the X-ray system was performed forward toward the lips, back to the pharyngeal wall, up to the nasal cavity, and downward to the upper esophageal sphincter, obtaining a side VF recording of 30 frames per second. The data were recorded on a DVD. Three blinded dentists (A Hiraoka, A Haruta, and MY) with specialized experience in evaluating videofluorographic recordings and established protocols following training on VF assessment determined the presence or absence of laryngeal penetration/aspiration, and clearance or prevalence of oral cavity residue, vallecular residue, or pharyngeal residue after one swallow. We also performed a semi-quantitative evaluation of oral cavity, vallecular, and pharyngeal residues, which were scored as grade 0 (no residue), grade 1 (thin coating of residue), or grade 2 (obvious residue). Aspiration/laryngeal penetration was categorized as grade 0 (none), grade 1 (laryngeal penetration), or grade 2 (aspiration beyond the vocal cords). In addition, the passage time of each anatomical landmark was measured and temporal analysis was performed. We calculated pharyngeal transit time (PTT), laryngeal elevation delay time (LEDT), and pharyngeal delay time (PDT). PTT is defined as the time from when the bolus tip reaches the lower border of the mandible to the complete passage of the bolus tail through the esophageal inlet (normal range 0.43–1.11 s) (19). LEDT is defined as the time from when the bolus tip reaches the vallecula to the peak of laryngeal elevation, in which the favorable cut-off value is 0.32 s (20). PDT is defined as the time from when the bolus tip reaches the intersection of the lower border of the mandible and the base of the tongue to the initiation of laryngeal elevation, in which the duration among healthy adults is 0–0.2 s (21). Furthermore, we evaluated the

TABLE 1 Patient backgrounds and indicators related to swallowing function.

	$n = 25$
Age, years	72.0 ± 5.9
Sex (female), *n* (%)	9 (36.0)
Duration, years	6 (1, 20)
Body mass index, kg/m^2	21.2 ± 2.8
Alcohol consumption, *n* (%)	2 (8.0)
Current smoking, *n* (%)	3 (12.0)
Hoehn & Yahr stage	3 (2, 4)
UPDRS score (total)	37 (19, 76)
UPDRS score (part 3)	23 (10, 50)
Dopa, mg	360 ± 203
LEDD, mg	583 ± 395
Maximum handgrip strength, kg	24.8 ± 5.6
Calf circumference, cm	33.8 ± 3.3
FOIS	7 (6, 7)
Tongue pressure, kPa	30.6 ± 8.5
VF findings	
Laryngeal penetration or Aspiration, *n* (%)	10 (40.0)
Oral cavity residue, *n* (%)	22 (88.0)
Epiglottic vallecula residue, *n* (%)	18 (72.0)
Pharyngeal residue, *n* (%)	15 (60.0)
Swallowing reflex delay, *n* (%)	4 (16.0)
Pharyngeal transit time, second	0.719 ± 0.122
Laryngeal elevation delay time, second	0.217 ± 0.146
Pharyngeal delay time, second	0.002 ± 0.134

UPDRS, Unified Parkinson's Disease Rating Scale; LEDD, Levodopa equivalent daily dose; FOIS, Functional Oral Intake Scale; VF, videofluoroscopic examination.
Data are expressed as mean ± standard deviation or median (minimum, maximum) for continuous variables and as frequencies and percentages for discrete variables.

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TABLE 2 Transition of indicators at baseline, 8, and 16 weeks from the start of the intervention.

	0 weeks (pre-intervention)	8 weeks (post-intervention)	*p*-value	16 weeks	*p*-value
Body mass index, kg/m^2	21.2 ± 2.8	21.3 ± 2.8	0.899	21.0 ± 2.5	0.782
UPDRS (total)	37 (19, 76)	42 (14, 78)	0.853	44 (15, 80)	0.554
UPDRS (part 3)	23 (10, 50)	25 (10, 51)	0.930	28 (11, 51)	0.669
Dopa, mg	360 ± 203	360 ± 203	1.000	360 ± 203	1.000
LEDD, mg	583 ± 395	583 ± 395	1.000	589 ± 392	0.957
Maximum handgrip strength, kg	24.8 ± 5.6	25.5 ± 5.4	0.665	26.7 ± 6.4	0.276
Calf circumference, cm	33.8 ± 3.3	33.8 ± 3.5	1.000	34.0 ± 3.2	0.830
FOIS	7 (6, 7)	7 (6, 7)	1.000	7 (6, 7)	1.000
Tongue pressure, kPa	30.6 ± 8.5	33.6 ± 8.0	0.199	34.6 ± 8.4	0.101
VF findings					
Laryngeal penetration or aspiration, *n* (%)	10 (40.0)	9 (36.0)	0.771	11 (44.0)	0.775
Oral cavity residue, *n* (%)	22 (88.0)	14 (56.0)	0.012*	19 (76.0)	0.270
Epiglottic vallecula residue, *n* (%)	18 (72.0)	17 (68.0)	0.758	15 (60.0)	0.371
Pharyngeal residue, *n* (%)	15 (60.0)	11 (44.0)	0.258	14 (56.0)	0.775
Swallowing reflex delay, *n* (%)	4 (16.0)	3 (12.0)	0.684	5 (20.0)	0.713
Pharyngeal transit time, s	0.719 ± 0.122	0.750 ± 0.167	0.461	0.755 ± 0.183	0.411
Laryngeal elevation delay time, s	0.217 ± 0.146	0.215 ± 0.189	0.967	0.256 ± 0.117	0.301
Pharyngeal delay time, s	0.002 ± 0.134	−0.020 ± 0.193	0.645	0.049 ± 0.142	0.238

UPDRS, Unified Parkinson's Disease Rating Scale; LEDD, Levodopa equivalent daily dose; FOIS, Functional Oral Intake Scale; EAT-10, Eating Assessment Tool-10; VF, videofluoroscopic examination.
Data are expressed as mean ± standard deviation or median (minimum, maximum) for continuous variables, and frequencies and percentages for discrete variables. Univariate analyses were performed compared to the baseline (0 weeks). *$p < 0.05$.

presence or absence of swallowing reflex delay, defined as liquid remaining in the pyriform sinuses for >0.1 s (3 frames) before swallowing (12). Three observers discussed their observations and reached a consensus for each observation or measurement.

2.4. Data acquisition

Clinical evaluation and diagnosis were conducted by two neurologists (MN and HY). The recorded data included body mass index, grip power, calf circumference, disease duration, alcohol drinking and smoking habits, Unified Parkinson's Disease Rating Scale score (22), medication, and Functional Oral Intake Scale score (23). Tongue pressure was assessed as previously described (24, 25). The levodopa equivalent daily dose (LEDD) was calculated based on a recent study (26). All evaluations were conducted in the ON state.

2.5. Sample size

We determined the necessary sample size based on initial assessments of coughing in individuals with neurodegenerative conditions. In these assessments, 28.6% of individuals exhibited a normal cough reflex following a 1% citric acid challenge. Assuming that 50% of individuals would demonstrate a normal cough reflex after 8 weeks of treatment, the estimated sample size was 27 participants. This calculation was made using an alpha level of 0.10, a power of 0.80, and accounting for a dropout rate of 10%.

2.6. Statistical analysis

The data are expressed as means ± standard deviation or medians (minimum, maximum) for continuous variables and as frequencies and percentages for discrete variables. Statistical analysis was performed using JMP statistical software, version 16 (SAS Institute Inc., Cary, NC, USA). To assess the efficacy of cervical percutaneous interferential current stimulation, we compared the VF results for each patient before the initial intervention with those obtained 8 weeks after the start or 8 weeks after the conclusion of the intervention (16 weeks from initiation). Additionally, we conducted a statistical comparison between the group that exhibited improvement and the group that did not. For the assessment of intergroup variances, appropriate statistical tests such as the $\chi 2$ test, Mann–Whitney *U*-test, or unpaired *t*-test were employed.

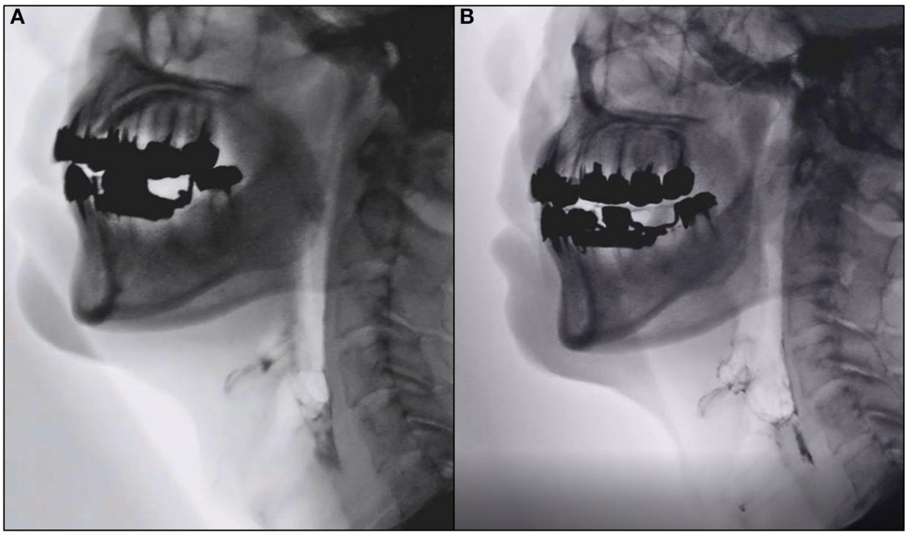

FIGURE 2
Representative images of videofluoroscopic examination. Images of videofluoroscopic examination from Case 5. **(A)** Pre-intervention (0 weeks). **(B)** Eight weeks post-intervention showing marked improvement in oral cavity residue.

The baseline data of patients were analyzed, and two-step strategies were applied to evaluate the relative importance of variables that exhibited improvement of VF findings using multiple logistic analysis. First, univariate analysis was performed. Subsequently, a multi-factorial analysis was performed with selected factors with a p-value <0.05 in univariate analysis. The correlation between factors was calculated using Pearson's correlation coefficients. We used basic factors (age, UPDRS total score tongue pressure). Statistical significance was set at $p < 0.05$.

3. Results

In this study, 27 participants were enrolled, and within 4 weeks after the start, two individuals withdrew their consent because of personal reasons. As a result, intervention and evaluation were conducted with 25 participants. The intervention was conducted without any deviations.

The patient demographics and swallowing-related indicator data at baseline (pre-intervention) are shown in Table 1. The laryngeal penetration/aspiration, oral cavity residue, epiglottic vallecula residue, and pharyngeal residue were found in remarkable frequency. Conversely, PTT, LEDT, and PDT were almost within the normal range. The number of patients who exhibited deviations from the standard values of LEDT and PDT delays was three and one, respectively. The number of swallowing reflex delays was 4 (16.0%).

The transitions of indicators at baseline, at the end of the intervention (8 weeks from the initiation of intervention), and at 8 weeks after the last intervention (16 weeks from the initiation of intervention) are shown in Table 2. At the intervention endpoint (8 weeks), oral cavity residue showed a significant improvement compared to that before the intervention (Figure 2). However, at 8 weeks after the intervention ended (16 weeks), no significant sustained improvement was observed. Additionally, no significant changes were observed in laryngeal penetration/aspiration, vallecular residue, and pharyngeal residue throughout the entire course when compared to the baseline. Moreover, no significant changes were found in PTT, LEDT, and PDT during the temporal analysis. Similarly, no significant change was observed in swallowing reflex delay. A semi-quantitative evaluation of aspiration/laryngeal penetration and oral cavity, vallecular, and pharyngeal residues was also performed, which demonstrated a significant improvement in the oral cavity residue at the intervention endpoint (8 weeks). However, no significant changes were observed in aspiration/laryngeal penetration or vallecular and pharyngeal residues (Supplementary Table 1).

At the baseline stage, there were 22 participants with oral cavity residue, and at the end of the 8-week intervention, nine patients showed improvement. Therefore, using the baseline data, a univariate analysis was conducted to examine the factors associated with improvement between these nine individuals and 13 who did not exhibit improvement. As a result, a higher body mass index and higher calf circumference were significantly associated with improved oral cavity residue ($p < 0.05$) (Table 3). These factors showed a strong correlation in Pearson correlation analysis, with a correlation coefficient of 0.782 ($p < 0.001$). Therefore, we conducted multivariate analysis by including age (Model 1), UPDRS total score (Model 2), and tongue pressure (Model 3), respectively, for each of body mass index and calf circumference to determine their validity as factors related to improving oral cavity residue. The results showed that body mass index and calf circumference were both significant correlating factors (Table 4). Conversely, three patients had no oral cavity residue at baseline, although one individual did at the end of the 8-week intervention. The disease duration, body mass index, calf circumference, and LEDD of the patient was 10 years, 17.5 kg/m^2, 32 cm, and 1,300 mg, respectively.

4. Discussion

In this study, we extensively investigated the swallowing disorders in patients with PD using the gold standard VF. We have previously conducted detailed examinations of swallowing disorders in patients with amyotrophic lateral sclerosis and stroke using similar methods (24, 27, 28). These diseases are generally neurologic disorders characterized by paralysis and muscle weakness. Additionally, a decrease in tongue pressure and oral phase impairments are the central aspects of swallowing disorders, with exceptions such as Wallenberg's syndrome (29). PD, on the other hand, primarily manifests as bradykinesia, without typical paralysis. Extrapolating knowledge from other neurological disorders to evaluate swallowing disorders in PD is impractical. Furthermore, our study results suggested that diverse factors contribute to swallowing disorders in patients with PD.

Laryngeal penetration/aspiration, oral cavity residue, epiglottic vallecula residue, and pharyngeal residue were observed at a significant frequency. Tongue pressure was well-maintained. Therefore, muscle weakness was probably not the cause. The lack of efficient motion is due to bradykinesia and muscle rigidity. Conversely, the temporal analysis of the swallowing reflex showed that it was mostly within the normal range. When comparing VF and brain lesion sites in patients with stroke, there is a correlation between delayed swallowing reflex initiation and basal ganglia lesions (12, 20). Considering that PD also involves abnormalities in the cerebral basal ganglia network, we anticipated the possibility of delayed swallowing reflex initiation. However, the results contradicted the expectations. Neurodegenerative diseases such as PD, in contrast to stroke, involve systematic disruptions in the nervous system, leading to distinct clinical manifestations. Moreover, the CPG for swallowing is located in the medulla near the nucleus ambiguous and solitary tract nucleus (30, 31). The onset of PD is associated with the dorsal motor nucleus of the vagus nerve, which is in a different location. This suggests that CPG impairment might be bypassed in PD.

In this study, cervical percutaneous interferential current stimulation significantly improved oral cavity residue. We speculate that sensory stimulation through Gentle Stim®–which assumes the stimulation of the glossopharyngeal and superior laryngeal nerves—may have activated the sensation and facilitated oral-phase initiation, as the posterior one-third of the tongue is controlled by the glossopharyngeal nerve. Furthermore, high body mass index and calf circumference were associated with improvement in oral cavity residue. Previous studies in healthy older individuals reported the positive correlation of body mass index and calf circumference with tongue pressure and tongue thickness (32). A thicker tongue reduces oral cavity volume and makes it more likely for tongue pressure to increase. A decrease in tongue pressure and

TABLE 3 Comparison between patients with and without improvement and non-improvement in oral cavity residue.

	Improved ($n = 9$)	Not improved ($n = 13$)	p-value
Age, years	71.1 ± 4.3	73.1 ± 7.4	0.481
Sex (female), n (%)	3 (33.3)	4 (30.8)	0.899
Duration, years	6 (3, 20)	10 (1, 13)	0.788
Body mass index, kg/m^2	23.9 ± 2.2	19.6 ± 1.8	<0.001*
Alcohol consumption, n (%)	0 (0)	1 (7.7)	0.394
Current smoking, n (%)	1 (11.1)	2 (15.4)	0.774
Hoehn & Yahr stage	3 (2, 4)	3 (2, 3)	0.616
UPDRS score (total)	36 (24, 76)	35 (19, 76)	0.738
UPDRS score (part 3)	23 (10, 50)	22 (14, 48)	0.920
Dopa, mg	356 ± 116	327 ± 219	0.724
LEDD, mg	587 ± 393	533 ± 373	0.745
Maximum handgrip strength, kg	27.3 ± 5.7	23.7 ± 5.5	0.148
Calf circumference, cm	36.3 ± 2.5	32.1 ± 3.1	0.003*
FOIS	7 (7, 7)	7 (6, 7)	0.460
Tongue pressure, kPa	32.7 ± 9.5	28.5 ± 6.7	0.237

UPDRS, Unified Parkinson's Disease Rating Scale; LEDD, Levodopa equivalent daily dose; FOIS, Functional Oral Intake Scale; EAT-10, Eating Assessment Tool-10; VF, videofluoroscopic examination.
Data are expressed as mean ± standard deviation or median (minimum, maximum) for continuous variables, and frequencies and percentages for discrete variables. $^*p < 0.05$.

TABLE 4 Multivariate analysis of oral cavity residue improvement.

	Model 1	p-value	Model 2	p-value	Model 3	p-value
	Odds ratio (95% CI)		Odds ratio (95% CI)		Odds ratio (95% CI)	
Body mass index	3.97 (1.16–13.54)	0.028*	3.83 (1.14–12.85)	0.030*	3.97 (1.16–13.58)	0.028*
Calf circumference	1.75 (1.12–2.74)	0.014*	1.73 (1.09–2.75)	0.020*	1.71 (1.17–3.05)	0.022*

CI, confidence interval.
Body mass index and calf circumference were identified as factors with $p < 0.05$ in univariate analyses for improvement in oral cavity residue. Multivariate analyses were performed using each factor and the basic factors; age (Model 1), Unified Parkinson's Disease Rating Scale total score (Model 2), and tongue pressure (Model 3), respectively.
$^*p < 0.05$.

tongue thickness has been associated with a decline in oral phase, namely, the passage of bolus from the oral cavity to the pharynx (24, 27). Based on these previous reports, it is consistent that body mass index and calf circumference are also involved in oral residue in patients with PD. Therefore, patients with maintained physical stature and muscle mass may potentially benefit more from cervical interferential current stimulation. However, further replication and investigation are essential to elucidate these factors conclusively in the context.

In addition to the cervical percutaneous interferential current stimulation used in this study, low-frequency neuromuscular electrical stimulation has also been reported to be a useful treatment for dysphagia (33). Low-frequency neuromuscular electrical stimulation primarily aims to induce muscle contractions, which can potentially contribute to the improvement of the muscle strength of swallowing-related muscles. In contrast, the interferential current stimulation used in the present study provides stimulation at levels that do not induce muscle contractions. Rather, this stimulation is characterized by interference waves reaching deep tissues and activating sensory nerves, resulting in less pain or discomfort caused by muscle contractions. As muscle strength, including tongue pressure, is relatively preserved in PD, it is important to select stimulation methods based on the pathophysiology of swallowing disorders. Future studies are needed to investigate how low-frequency neuromuscular electrical stimulation contributes to swallowing disorders in PD.

This study has several limitations. First, this study is a single-site single-group intervention trial. A randomized controlled trial including a non-intervention/sham stimulation group for intergroup comparison should be considered in future research. This study is the first investigation into the effectiveness of cervical interferential current stimulation among patients with PD. One of the objectives of this exploratory study was to explore factors that show improvement through the intervention of percutaneous interferential current stimulation. In the future, despite ethical challenges, intergroup comparison trials should be conducted. Second, one of the challenges of this exploratory study was the limited number of patients under investigation. This study focused on cervical interferential current stimulation as the primary endpoint to improve cough reflex testing. Therefore, the sample size was determined based on previous studies and existing literature. However, to comprehensively examine and analyze swallowing disorders in patients with PD and diverse symptoms, a substantial number of patients must be included for investigation. Therefore, further recruitment of patients is necessary for future research.

This exploratory study provided new insights into swallowing disorders in patients with PD. Additionally, the usefulness of cervical interferential current stimulation was partially demonstrated. Considering that PD causes diverse symptoms, further recruitment of patients and knowledge is warranted.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Certified Review Board at Hiroshima University. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

MN: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Validation, Writing—original draft. MT: Conceptualization, Data curation, Investigation, Methodology, Validation, Writing—original draft. HY: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Validation, Writing—original draft. YH: Data curation, Writing—original draft. AHa: Formal analysis, Investigation, Methodology, Writing—original draft. AHi: Formal analysis, Investigation, Methodology, Writing—original draft. MY: Investigation, Methodology, Validation, Writing—original draft. TN: Data curation, Investigation, Methodology, Writing—review & editing. KU: Conceptualization, Methodology, Writing—review & editing. KY: Data curation, Investigation, Methodology, Writing—original draft. YS: Conceptualization, Methodology, Writing—review & editing. YM: Project administration, Supervision, Validation, Writing—review & editing. HM: Conceptualization, Supervision, Validation, Writing—review & editing.

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Conflict of interest

HM received honoraria from Eisai, Shionogi, Otsuka Pharmaceutical, Sumitomo Pharma.

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Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fneur.2023.1279161/full#supplementary-material

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Quality appraisal of clinical practice guidelines for the management of Dysphagia after acute stroke

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Objectives: Dysphagia is a common complication in stroke patients, widely affecting recovery and quality of life after stroke. The objective of this systematic review is to identify the gaps that between evidence and practice by critically assessing the quality of clinical practice guidelines (CPGs) for management of dysphagia in stroke.

Methods: We systematically searched academic databases and guideline repositories between January 1, 2014, and August 1, 2023. The Appraisal of Guidelines for Research and Evaluation (AGREE II) instrument was used by two authors to independently assess CPG quality.

Results: In a total of 14 CPGs included, we identified that three CPGs obtained a final evaluation of "high quality," nine CPGs achieved "moderate quality" and two CPGs received "low quality." The domain of "scope and purpose" achieved the highest mean score (91.1%) and the highest median (IQR) of 91.7% (86.1, 94.4%), while the domain of "applicability" received the lowest mean score (55.8%) and the lowest median (IQR) of 55.4% (43.2, 75.5%).

Conclusion: The CPG development group should pay more attention to improving the methodological quality according to the AGREE II instrument, especially in the domain of "applicability" and "stakeholder involvement;" and each item should be refined as much as possible.

KEYWORDS

stroke, dysphagia, clinical practice guidelines, AGREE II, quality appraisal

1 Introduction

Globally, stroke remained the second-leading cause of death and the third-leading cause of death and disability combined in 2019 (1, 2). Dysphagia is a common complication in stroke patients, widely affecting recovery and quality of life after stroke and increasing mortality risk through increased risk of dehydration, malnutrition and pneumonia (3). The incidence of dysphagia varies widely depending on the method of assessment, compared with clinical assessment (30–55%) and video rheology (64–78%), a lower incidence was detected using initial screening tools (37–43%) (4). However, managing dysphagia correctly and effectively can shorten hospital stays, reduce the risk of death, and decrease healthcare costs (5, 6).

Clinical practice guidelines (CPGs) are a type of declaration that include evidence-informed recommendations aimed at optimizing patient care that are informed by a systematic review of evidence and an assessment of the benefits and harms of alternative care options (4). To date, a number of CPGs have been developed and updated with the aim of ensuring optimal dysphagia management of stroke patients. CPGs would contribute to improving the quality of health care, for example, providing evidence for clinicians to make decisions about patient care and determining appropriate medical criteria, thereby identifying gaps between evidence and practice (7). Nevertheless, hospital personnel adherence to evidence-based stroke care is limited (8), translating evidence into clinical practice is challenging, and implementation of these CPGs in clinical practice remains suboptimal (9, 10).

The quality of the CPGs has a direct impact on utilization (11, 12), and the purpose of this study was to assess the quality of guidelines for managing poststroke dysphagia. Therefore, we used the Appraisal of Guidelines for Research and Evaluation II (AGREE II) instrument (13) to evaluate the quality of CPGs for dysphagia management after stroke, which may be helpful in identifying the potential factors that impact the quality of CPGs. The findings would illustrate the gaps between evidence-based guidelines and clinical practice and attempt to explore potential measures of improvement.

2 Materials and methods

2.1 Search strategy

A comprehensive literature search was conducted by two authors to identify CPGs for the prevention, diagnosis, and treatment of dysphagia after acute stroke between January 1, 2014, and August 1, 2023. The following databases were searched: PubMed, Web of Science and EMBASE, Clinical Practice Guidelines, the National Institute for Health and Care Excellence, National Guideline Clearinghouse, World Health Organization, Scottish Intercollegiate Guideline Network, New Zealand Guidelines Group and BMJ Best Practice. Search strategies were tailored according to each database (The specific search strategy is displayed in Supplementary File 1). All results were imported into EndNote (Version.X9.2), where duplicates were removed. A third author resolved any disagreements.

2.2 Eligibility criteria

The inclusion criteria were as follows: (1) International and national CPGs published on the management of dysphagia after acute stroke; (2) Published or updated from January 1, 2014 to August 1, 2023; (3) Published in English; and (4) Guidelines focused on adult patients. The excluded criteria were as follows: (1) Guideline-related interpretation, application evaluation or brief versions, etc.; (2) Full text not available; and (3) Guidelines under development or withdrawal.

2.3 Data screening and extraction

The titles and abstracts of all search results were screened by two authors before checking the full text. In addition, two authors scanned the reference lists of the confirmed papers to identify more relevant CGPs. Then, they extracted the characteristics of the CPGs including year, developer, grading system, country/region, target population, and multidisciplinary team using a predesigned standardized data extraction form.

2.4 Quality assessment

The quality of the 14 CPGs was appraised by two authors trained using the AGREE II instrument, which is a reliable tool that is widely used to assess the quality of CPGs (13). AGREE II consists of 23 items organized into six domains and two overall assessment portions. Each item was scored from 1 to 7 (1 = strongly disagree, 7 = strongly agree). Prior to the formal assessment, we discussed the assessment criteria based on the AGREE II manual and training tools to maintain a consistent understanding of each item. After scoring, we organized the CPGs and randomly cross-checked 10% (14) to ensure consistency between authors, especially for items with wide variations in scoring.

The standardized scores for each domain were computed based on the achievement scores (13), as follows: The maximum possible score of domain = 7 (strongly agree) × number of items × number of evaluators; a minimum possible score of domain = 1 (strongly disagree) × the number of items x the number of evaluators. The standardized scores = (obtained score − minimum possible score)/(maximum possible score − minimum possible score) × 100%.

The AGREE II manual does not offer any advice on how to explain the scores. In accordance with previous studies (15, 16), if a CPG scored above 70% on six domains, it was classified as 'high quality'; if a CPG scored above 70% on three to five domains, it was classified as 'moderate quality'; and if a CPG scored less than 70% on ≥ two domains, it was classified as 'low quality'.

2.5 Statistical analysis

All data were analyzed using IBM SPSS Statistics Version 26.0 software and Microsoft Excel 2021. Mean, median and interquartile range (IQR) were computed for the domain scores. The intraclass correlation coefficient (ICC) was computed to measure the interrater agreement when performing a quality appraisal of the CPGs among the two appraisers to ensure the reliability of our conclusions. The level of ICC was classified according to commonly cited cutoffs: poor (< 0.50), fair (0.50–0.75), good (0.75–0.90) or excellent (0.90–1.00) (17).

3 Results

A total of 1,208 titles and abstracts were generated through database and manual searches. After deleting duplicates, 730 articles were filtered by title and abstract. A total of 36 full-text CPGs were screened for eligibility, and 14 CPGs were included in our systematic review. Figure 1 provides the PRISMA flow chart (18). Table 1 shows the general characteristics of the CPGs included in the analysis. Regarding geographical distribution, six of them are from Europe, the US and Canada all have two CPGs each, while Brazil, China, Turkey, Australia and New Zealand have only one.

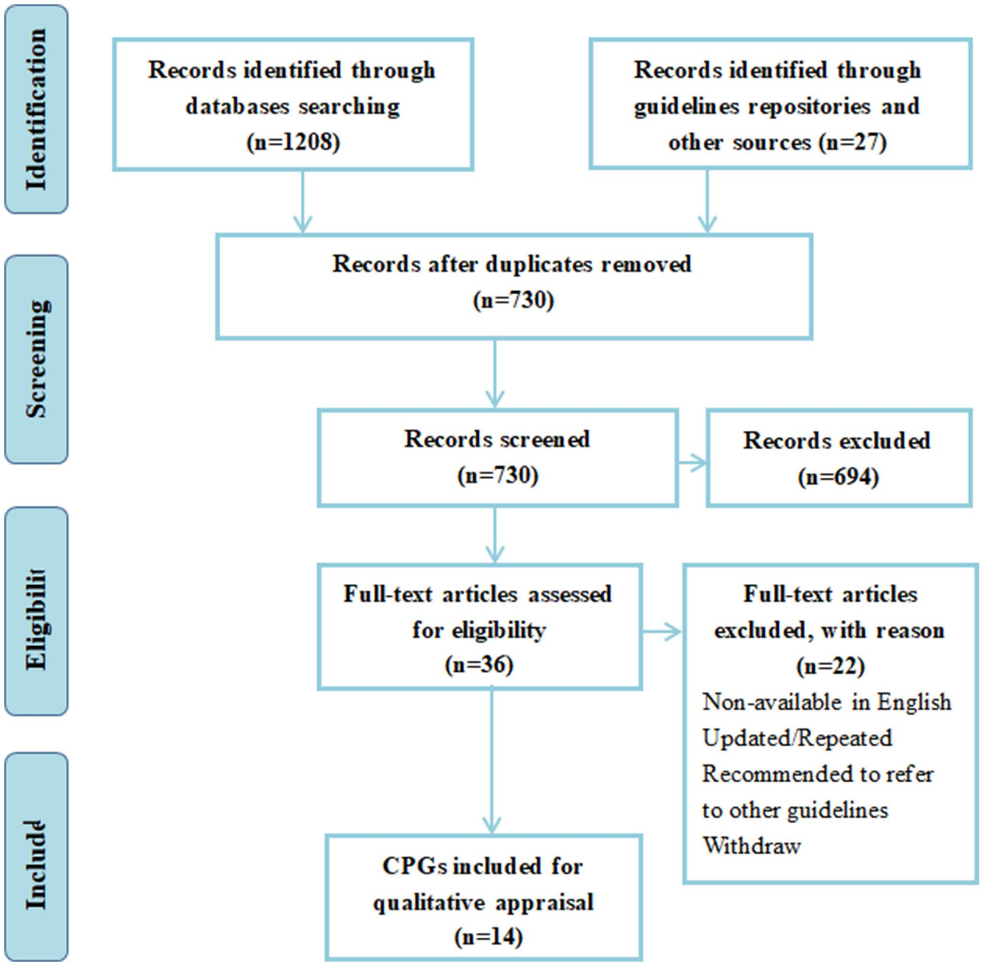

FIGURE 1
PRISMA flow chart of searching and selecting guidelines.

3.1 Quality of CPGs according to the AGREE II domains

Table 2 reports the ICC score, overall quality and recommendation comments of all CPGs. In total, 14 CPGs were included, only three CPGs were found to be of high quality with all domains reaching a score higher than 70%, nine CPGs were graded as moderate quality and the remaining two were classified as low quality. The evaluation results of the two appraisers were reliably consistent, with ICCs (95% CI) ranging from 0.75 (0.48, 0.88) to 0.90 (0.82, 0.99).

The quality of CPGs evaluated by AGREE II varied widely, not only between guidelines, but also between domains within guidelines. Figure 2 shows the score distribution of the 6 domains among the 14 CPGs. Figure 3 shows the mean score of each domain for all CPGs sorted by quality classification.

3.1.1 Scope and purpose

The domain of "scope and purpose" obtained the highest mean (91.1%) and the highest median (IQR) score of 91.7% (86.1, 94.4%). Moreover, all guidelines achieved over 70% in this domain, but only two of them had a maximum score of 100% (22, 29).

3.1.2 Stakeholder involvement

The standardized scores in this domain ranged from 38.9 to 91.7%, with nine of 14 CPGs scoring above 70%. Most of the poor scores are due to the views and preferences of the target population (patients, public, etc.) have not been sought (22, 32).

3.1.3 Rigor of development

Regarding the standardized scores in this domain, the mean was 70.5%, and the median (IQR) was 75.0% (62.2, 81.3%). GSN2021 (32) obtained the lowest scores (43.8%). Most CPGs lacked clarity in describing all stages of the methodological development or did not provide a procedure for updating the guidelines.

3.1.4 Clarity of presentation

In this domain, the mean score was 87.3%, and the median (IQR) was 90.3% (79.5, 97.2%). Six CPGs received above 90%. In contrast, GSN2021 (32) obtained the lowest score of 63.9%, which means that the guideline development group did not present recommendations clearly.

3.1.5 Applicability

This domain yielded the lowest mean score of 56.8% and the lowest median (IQR) score of 55.4% (43.2, 75.5%). Five CPGs

TABLE 1 General characteristics of the CPGs included in the analysis.

No.	Year	Developer	Country/Region	Grading system	Evidence based	Intended population	Multidisciplinary team
1	2022	BAN (19)	Brazilian	Evidence: A, B,C Recomm: Class I-III	Clinical trials, meta-analyses, and systematic reviews	Health professionals	Not reported
2	2020	IMSWT (20)	China	Evidence: 1–4 Recomm: GRADE	Existing guidelines and systematic reviews	Medical practitioners, including Chinese herbal medicine specialists, acupuncturists, integrative medicine practitioners, physicians, physical therapists, and clinical pharmacists	Traditional Chinese medicine, integrative medicine, neurology, neurovascular intervention, neurosurgery, emergency neurology, rehabilitation, acupuncture, nursing, pharmacy, evidence-based medicine, and standardization of Chinese medicine and health economics.
3	2022	PMR (21)	Turkey	Evidence: 0–10 Recomm: OC, AC, OD	A 3-round Delphi questionnaire/ survey, expert consensus	Not reported	4 physical medicine and rehabilitation medical doctors, consultant experts
4	2016	NICE (22)	UK	Not reported	Not reported	Healthcare professionals, Commissioners and providers of services, People who have had a stroke, their families and carers	Not reported
5	2021	ESO (23)	Europe	Evidence: high, moderate, low, very low Recomm: Strong/Weak	Systematic reviews, meta-analysis, RCTs	Physicians, speech-and-language therapists as well as stroke-nurses, and all the members of the multidisciplinary team	a phoniatrician, a surgeon, two neurologists, a geriatrician, a gastroenterologist, a stroke physician, a pharmacist and a rehabilitation physician
6	2021	ESPEN (24)	Europe	Evidence: 1–4 Recomm: A, B,0,GPP	Not reported	Hospitals, rehabilitation centers, and nursing homes	Six physicians and five dietitians
7	2018	ESPEN (25)	Europe	Evidence: 1–4 Recomm: A, B,0,GPP	Systematic reviews and meta-analysis	Patients with dysphagia and malnutrition	Clinical nutrition, Neurology, Geriatrics, Dietetics and Intensive Care
8	2017	NSF (26)	AN	Recomm: Weak, Strong	Systematic reviews and RCTs	Healthcare professionals	Clinical expert, people with relevant lived experience
9	2016	RCP (27)	UK	Not reported	All high-quality evidence available	Clinicians, patients and their families and carers, and those with responsibility for commissioning stroke services	Clinicians, people with stroke and their families
10	2016	AHA/ASA (28)	USA	Evidence: A, B, C Recomm: Class I-III	Not reported	The members of the multidisciplinary team	Stroke patient, caregivers, physicians, nurses, occupational therapists, recreation therapists, nutritionists, social workers,
11	2019	AHA/ASA (29)	USA	Evidence: A, B, C Recomm: Class I-III	Existing systematic reviews, meta-analysis and RCTs	Prehospital care providers, physicians, allied health professionals, and hospital administrators	Not reported
12	2020	CSA (30)	Canada	Evidence: A, B, C Recomm: Not reported	Systematic reviews, meta-analyses, RCTs, and observational studies	People who have already had a moderately or severely disabling stroke	Stroke neurologists, a geriatric psychiatrist, a clinical pharmacologist, neuropsychologists, physiotherapists, occupational therapists, a speech-language pathologist, nurses,

(Continued)

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TABLE 1 (Continued)

No.	Year	Developer	Country/Region	Grading system	Evidence based	Intended population	Multidisciplinary team
13	2022	CSA (31)	Canada	Evidence: high, moderate, low, Recomm: Strong/ Weak	Systematic reviews, meta-analyses, RCTs, and observational studies	All healthcare providers, health system leaders and planners, and people living with stroke	Seven people with stroke and one caregiver
14	2021	GSN (32)	German	Not reported	RCTs, cohort studies, systematic meta-analysis, and guideline publications	Not reported	Dysphagia experts from 27 medical societies

BAN, Brazilian Academy of Neurology; IMSWT, Integrative Medicine for Stroke working team; GPP, good practice points; GRADE, The Grading of Recommendations Assessment, Development and Evaluation; PMR, Turkish Society of Physical Medicine and Rehabilitation; WGO, World Gastroenterology Organization; CPGs, clinical practice guidelines; NICE, National Institute for Health and Care Excellence; ESO, European Stroke Organization; ESPEN, The European Society for Clinical Nutrition and Metabolism; NSF, National Stroke Foundation; RCP, Royal College of Physicians; AHA/ASA, American Heart Association/American Stroke Association; CSA, Canadian Stroke Association; AN, Australian and New Zealand; GSN, German Society of Neurology.

TABLE 2 Appraisal of Guidelines for Research and Evaluation (AGREE) II version result for clinical practice guidelines.

CPG	Domain1	Domain2	Domain3	Domain4	Domain5	Domain6	ICC (95% CI)	Overall quality	Recomm Comment
BAN2022 (19)	91.7	66.7	80.2	91.7	75.0	83.3	0.86(0.74–0.95)	Moderate	YES*
NICE2022 (22)	100	38.9	52.1	80.0	64.7	62.5	0.82(0.62–0.92)	Low	NO
IMSWT2020 (20)	94.4	91.7	85.4	88.9	48.0	91.7	0.77(0.58–0.85)	Moderate	YES*
ESO2021 (23)	91.7	86.1	75.0	97.2	22.9	100	0.80(0.58–0.91)	Moderate	YES*
ESPEN2018 (25)	91.7	62.4	65.6	91.7	45.8	95.8	0.75(0.48–0.88)	Moderate	YES*
ESPEN2021 (24)	86.1	72.2	67.7	77.8	54.2	87.5	0.90(0.82–0.99)	Moderate	YES*
NSF2017 (26)	86.1	88.9	67.7	91.7	56.5	100	0.79(0.57–0.91)	Moderate	YES*
PMR2022 (21)	80.6	86.1	50.0	83.3	52.1	83.3	0.87(0.64–0.96)	Moderate	YES*
RCP2016 (27)	91.7	88.9	84.4	97.2	77.0	100	0.75(0.49–0.89)	High	YES
AHA/ASA2016 (28)	94.4	87.4	85.4	100	79.2	87.5	0.78(0.54–0.90)	High	YES
AHA/ASA2019 (29)	100	61.1	78.1	77.8	32.9	83.3	0.85(0.53–0.94)	Moderate	YES*
CSA2020 (30)	88.9	66.7	75.0	83.3	70.8	87.5	0.84(0.68–0.95)	Moderate	YES*
CSA2022 (31)	91.7	86.1	76.0	97.2	81.3	100	0.78(0.54–0.90)	High	YES
GSN2021 (32)	86.1	56.8	43.8	63.9	35.4	100	0.80(0.55–0.91)	Low	NO
Mean	91.1	74.3	70.5	87.3	56.8	90.2			
Median (IQR)	91.7(86.1–94.4)	79.2(62.1–87.8)	75.0(62.2–81.3)	90.3(79.5–97.2)	55.4(43.2–75.5)	89.6(83.3–100)			

YES*, recommended with modifications.

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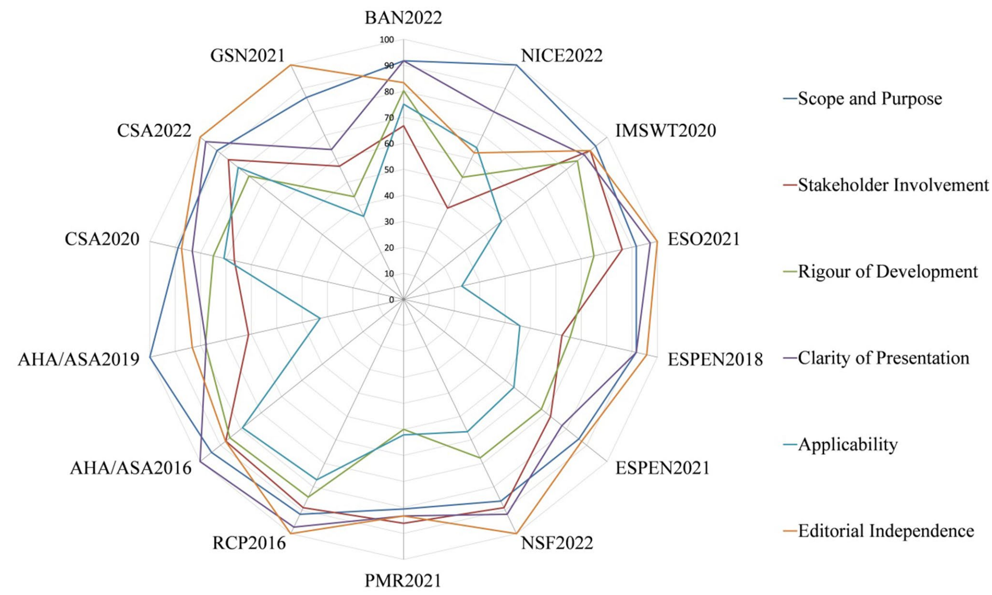

FIGURE 2
Score distribution of the six domains among the 14 CPGs.

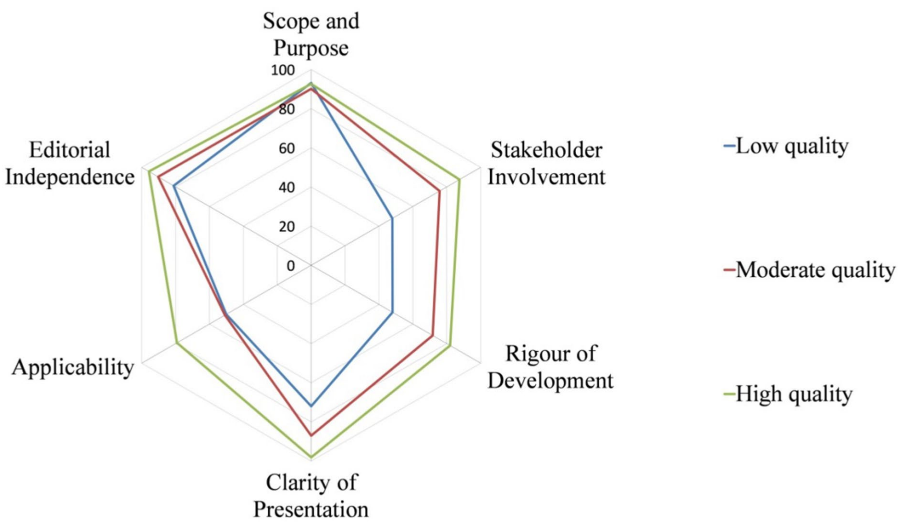

FIGURE 3
Mean score of each domain for all CPGs sorted by quality classification.

scored more than 70% (19, 27–29, 31), but only CSA2022 (31) scored above 80%, whereas the other guidelines described certain items in the domain unsatisfactorily.

3.1.6 Editorial independence

In this domain, the mean was 90.2%, and the median (IQR) was 89.6% (83.3, 100%). Five CPGs (23, 26, 27, 31, 32) received full marks in this domain, with the exception of NICE2022 (22), which did not explicitly provide information on editorial independence and the competing interests of members of the CPG development group have not been recorded and addressed.

4 Discussion

The present study proposes a critical review that evaluates the quality of 14 CPGs developed to manage dysphagia in acute stroke using the AGREE II tool (13). Depending on our results, the quality of CPGs evaluated by AGREE II varied significantly, not only between guidelines, but also between domains within guidelines. RCP2016 (27), AHA/ASA2016 (28) and CSA2022 (31) were classified as high quality and thus were recommended based on the AGREE II tool. Among the domains, "scope and purpose" obtained the highest mean score of 91.1% and the highest median (IQR) score of 91.7% (86.1, 94.4%), while "applicability" yielded the lowest mean score of 56.8% and the lowest median (IQR) score of 55.4% (43.2, 75.5%).

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Based on the AGREE II reported items, the domain of "applicability" performed the worst, which is consistent with other quality assessment results of CPGs in different healthcare topics (33, 34). Many CPGs failed to identify and describe the potential facilitators, barriers and advice or tools on how the recommendations can be put into practice. This may be one of the reasons why clinical implementation is not as effective as it could be (9, 35). To address this issue, we find that implementation science approaches are feasible, and a quality improvement intervention that includes online educational videos, mobile health technology, simplified versions of the guidelines manual, audits and feedback, is recommended to improve the CPG adherence of medical staff and patients, user awareness and CPG uptake (36–39).

Regarding the domain of "stakeholder involvement," some CPGs did not clearly describe the guideline development group or the views and preferences of the target population (patients, public, etc.) were not been sought. During the development of CPGs, patients and a variety of stakeholders, such as clinicians of all types, insurance payers and funders, health policy decision makers, and experts should be involved in the development of CPGs to set priorities, ensure feasibility, and promote distribution and compliance (6, 40, 41).

Most of the CPGs lacked clarity in describing the crucial stages of the methodological development, especially in external review and procedure for updating, which is important for transparency and applicability (42). In addition, guidelines would benefit from a more prescriptive and standardized evidence-based approach to developing recommendations and avoiding the use of ambiguous recommendations. Two of the included CPGs [IMSWT2020 (20), CSA2022 (31)] used the AGREE II tool during the external review and development phase. Although IMSWT2020 (20) used the AGREE II instrument, high quality is still not achieved in the domain of applicability. Therefore, the AGREE II instrument should be considered in the process of planning, developing and publishing CPGs for guideline development groups (13). Our results were largely similar to the results of CPG quality appraisal in different clinical topics (43–45), indicating that the problems in CPG development have some commonality. The CPG development group should pay more attention to improving the methodological quality according to the AGREE II instrument, and each item should be refined as much as possible (16, 42).

In addition to focusing on improving the transparency and methodological rigor of the guideline development process, the quality of guidelines is more dependent on high-quality evidence. However, most of the recommendations in the above guidelines are based on low to moderate quality evidence, and even some of them are not based on evidence. More high-quality evidence is needed for the management of post-stroke dysphagia, such as how to select instruments to evaluate swallowing with sensory tests (29), rational dietary programs (24), and effective therapies (31), which are extremely important for improving the quality of care for patients with post-stroke dysphagia.

Our study has several strengths. First, before the formal assessment, two assessors discussed the appraisal criteria according to the AGREE II manual and training tools to maintain the understanding of each item in line with each other. After scoring, the CPGs were collated with a randomized 10% cross-check (14) to ensure consistency between authors, especially for the items with significantly different scores. Furthermore, to the best of our knowledge, this is the first study that compares and evaluates the quality of CPGs in the nutritional management of stroke patients.

Due to language or publication restrictions, our review is limited to CPGs written in English, and excluding CPGs written in other languages may introduce bias. Furthermore, AGREE II does not provide an explicit cutoff to distinguish between high quality, moderate quality, and low quality CPGs. We defined them based on previous studies, but we are not exempt from misinterpretation that may derive from heterogeneity in the formulation and wording of recommendations. In addition, it is worth noting that in this study, only the critical appraisal of the quality development of the guidelines was performed, without any assessment of the quality of the guidelines' content.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

S-LG: Data curation, Methodology, Writing – original draft, Writing – review & editing. C-QL: Methodology, Writing – review & editing. Q-HH: Data curation, Writing – review & editing. X-RD: Data curation, Writing – review & editing. Y-WL: Supervision, Validation, Writing – review & editing, Methodology. KL: Supervision, Validation, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fneur.2023.1310133/full#supplementary-material

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Research trends and hotspots of post-stroke dysphagia rehabilitation: a bibliometric study and visualization analysis

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Background: Post-stroke dysphagia (PSD) is one of the most prevalent stroke sequelae, affecting stroke patients' prognosis, rehabilitation results, and quality of life while posing a significant cost burden. Although studies have been undertaken to characterize the pathophysiology, epidemiology, and risk factors of post-stroke dysphagia, there is still a paucity of research trends and hotspots on this subject. The purpose of this study was to create a visual knowledge map based on bibliometric analysis that identifies research hotspots and predicts future research trends.

Methods: We searched the Web of Science Core Collection for material on PSD rehabilitation research from its inception until July 27, 2023. We used CiteSpace, VOSviewer, and Bibliometrix R software packages to evaluate the annual number of publications, nations, institutions, journals, authors, references, and keywords to describe present research hotspots and prospective research orientations.

Results: This analysis comprised 1,097 articles from 3,706 institutions, 374 journals, and 239 countries or regions. The United States had the most publications (215 articles), and it is the most influential country on the subject. "Dysphagia" was the most published journal (100 articles) and the most referenced journal (4,606 citations). Highly cited references focused on the pathophysiology and neuroplasticity mechanisms of PSD, therapeutic modalities, rehabilitation tactics, and complications prevention. There was a strong correlation between the terms "validity" and "noninvasive," which were the strongest terms in PSD rehabilitation research. The most significant words in PSD rehabilitation research were "validity" and "noninvasive brain stimulation," which are considered two of the most relevant hotspots in the field.

Conclusion: We reviewed the research in the field of PSD rehabilitation using bibliometrics to identify research hotspots and cutting-edge trends in the field, primarily including the pathogenesis and neurological plasticity mechanisms of PSD, complications, swallowing screening and assessment methods, and swallowing rehabilitation modalities, and this paper can provide in the follow-up research in the field of PSD rehabilitation. The results of this study can provide insightful data for subsequent studies in the field of PSD rehabilitation.

KEYWORDS

stroke, deglutition disorders, bibliometrics, rehabilitation, CiteSpace, VOSviewer

1 Introduction

According to a 2019 Global Burden of Disease Study research, stroke is still the second largest cause of mortality (11.6% of total deaths) and the third major cause of disability (1). Post-stroke dysphagia (PSD) is the most prevalent post-stroke complication, occurring between 37 and 78% of the time, and is difficult to recover from during a stroke (2). In accordance with studies, patients with dysphagia have a higher risk of aspiration and pneumonia than non-dysphagic patients (3, 4), resulting in lengthier hospitalization and an increased risk of death (5). Furthermore, qualitative research revealed that dysphagia has a negative impact on patients' mental health, social communication, and family roles, in addition to modifying their physical functioning and eating habits (6). Early discovery, evaluation, and rehabilitation can lower the risk of hypoxia, lung infection, malnutrition, and other complications (7), Early screening is thus the first step in dysphagia rehabilitation; however, many clinical swallowing screens have high sensitivity but low specificity, and the accuracy of bedside swallowing screening tools to identify dysphagia in the acute phase of stroke remains unknown (8).

Neurostimulation of the pharyngeal motor cortex is the focus of neurological rehabilitation of swallowing function, with techniques separated into peripheral sensory and central stimulation (9). The use of drugs such as transient receptor potential (TRP) channel agonists and retropharyngeal or transcutaneous electrical stimulation (TES) methods to boost sensory input to improve oropharyngeal swallowing responses is referred to as peripheral stimulation. Transcranial direct current stimulation (tDCS) and repetitive transcranial magnetic stimulation (rTMS) are among the more frequently utilized modalities of central stimulation for the treatment of PSD (10, 11). Notably, speech and language pathology have significance in the rehabilitation of individuals with dysphagia (12).

Bibliometric analysis is useful in defining the current state of many research fields, as well as the scientific accomplishments of researchers, institutions, and countries, as well as potential research hotspots (13). Bibliometric analyses of global healthy eating, ophthalmology, and the application of artificial intelligence in diabetic retina by a number of researchers have offered strong insights into study topics of interest (14–16). The rehabilitation of PSD has garnered extensive attention in recent years, however, the research hotspots and upcoming research paths have yet to be clarified. As a result, we conducted a bibliometric analysis of publications on PSD rehabilitation from the Web of Science Core Collection (WoSCC) database during the last 25 years. Bibliometric analysis software is used to explore countries, institutions, journals, authors, references, and keywords, to learn about countries, journals, institutions, and authors with top influence in the research field; to acquire about burning references and hot keywords, to form a clustering theme of research in the field of PSD rehabilitation, to create understandable visual models and analyze current research trends and hotspots to reveal the evolution and development of the research area and anticipate future research directions. This study not only provides authors and researchers with an overall visual knowledge map and significant insights into the topic of PSD rehabilitation, but it additionally provides meaningful references for future research.

2 Materials and methods

2.1 Search strategy and data collection

Web of Science Core Collection (WoSCC) is a globally influential, authoritative, and comprehensive database that provides comprehensive citation data covering multiple disciplines and has a unique advantage in conducting multidisciplinary and international bibliometric analyses (17); thus, we chose WoSCC as the source database for data retrieval in this research. The data retrieval strategy is as follows: TS = (stroke OR apoplexy OR "cerebrovascular accident" OR "brain vascular accident" OR "cerebral hemorrhage" OR encephalorrhagia OR "cerebral ischemia") AND TS = ("Deglutition Disorder" OR "Swallowing Disorder*" OR Dysphagia OR "Oropharyngeal Dysphagia" OR "Esophageal Dysphagia") AND TS = (rehabilitation OR recovery), The search period was set to July 27, 2023, and the genre of material was limited to articles and reviews. Irrelevant material was eliminated by skimming the titles and abstracts, which included Proceeding Papers, Meeting Abstracts, Letters, and so on. Finally, 1,097 articles were found to satisfy the inclusion criteria. The flow chart of the literature screening is shown in Figure 1.

2.2 Data analysis

We selected articles published within the last 25 years so that we could not only analyze the literature's development cycle and gain insight into how the impact of the literature has changed over time, which would be useful for researchers interested in the field of PSD rehabilitation to understand the history of the field and its evolutionary trends but also identify the literature that has had a key impact on the field of study at each stage of its development and authors. We also uncover research hotspots and trends in the field of PSD rehabilitation by using bibliometric analysis algorithms to find references with the highest citation bursts, keywords, and keyword clustering.

The steps for bibliometric analysis are as follows: We used VOSviewer 1.6.19 (Leiden University, Netherlands), CiteSpace 6.1.R6 (Drexel University, PA, United States), and the Bibliometrix R package (4.1.3) to analyze the included literature. The main contents of the analysis include countries, institutions, journals, authors, references, and keywords. Data was processed using Excel.

3 Results

3.1 Analysis of publication outputs

The analysis contains 1,097 papers published between 1998 and 2023, containing 885 articles and 212 reviews. The total h-index is 69, the total number of citations is 24,141, and the total number of frequently referenced articles is 21.99. Figure 2 depicts the annual publication and citation counts for PSD rehabilitation, which shows a generally consistent but unstable growth trend, with a slow increase in the number of publications between 1998 and 2013, ranging from about 10 to 40 articles per year, and then an accelerated growth beginning after 2014, except a slight decline in the number of publications in 2015 and 2021. The overall trend is upward and will

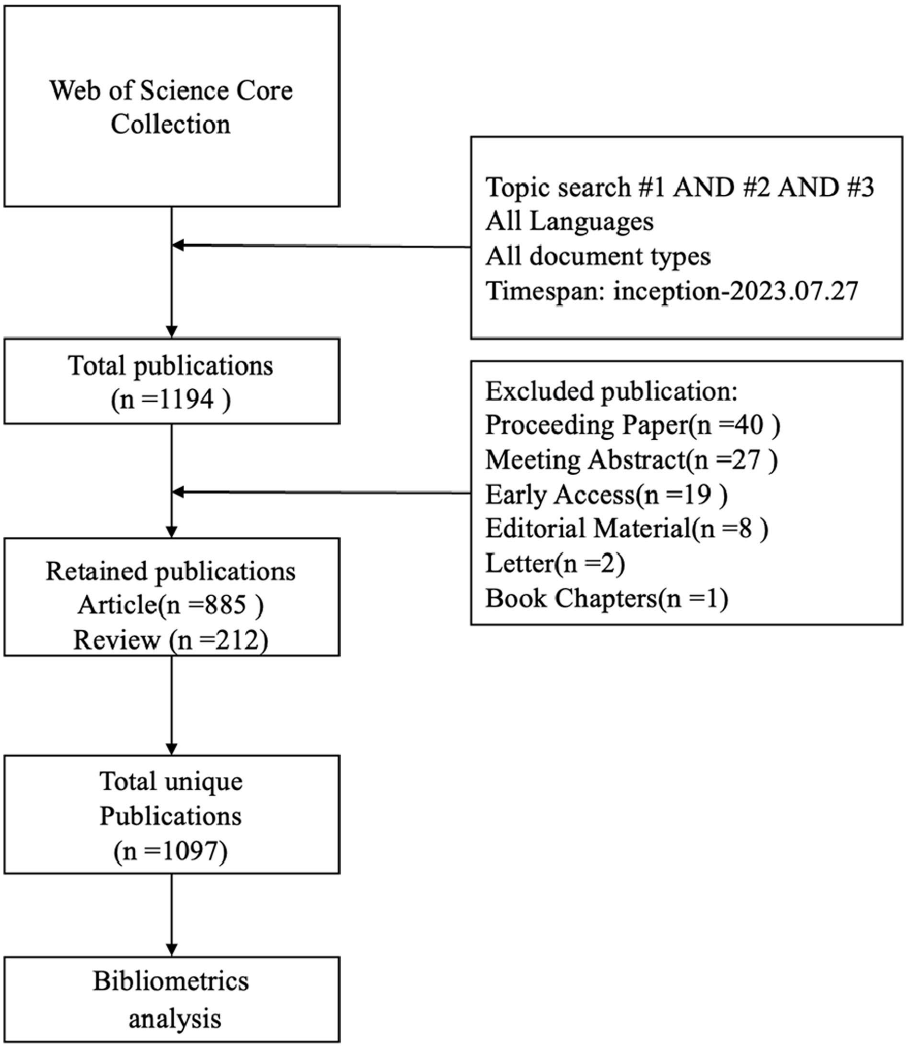


FIGURE 1
Flow chart of literature selection.

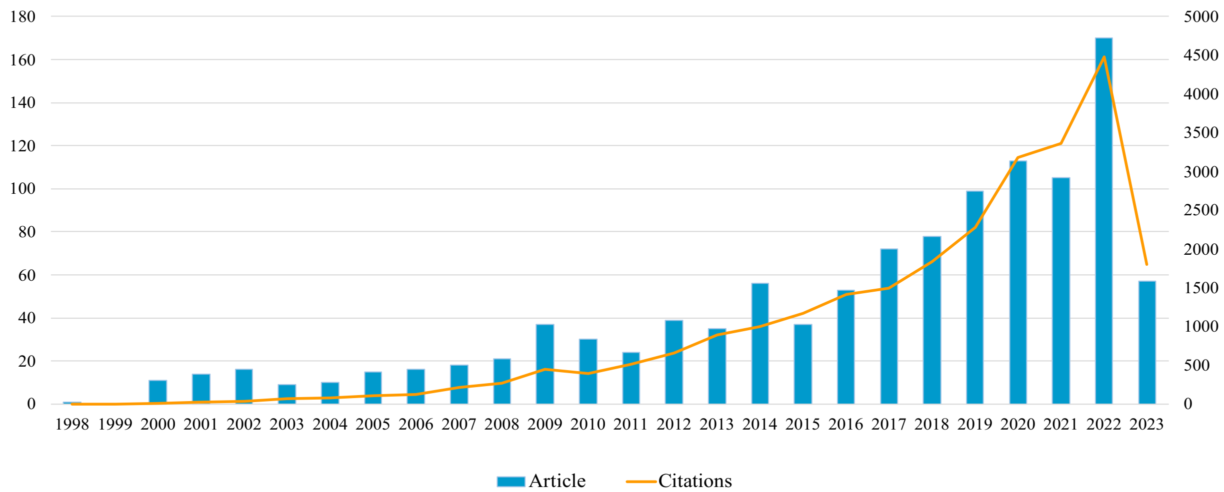


FIGURE 2
Annual publication trend of PSD rehabilitation.

peak in 2022, indicating that researchers are focusing more on PSD rehabilitation.

3.2 Country/region and institution contributions

PSD rehabilitation research is being conducted in 239 countries/regions by 3,706 institutions. The top 10 countries and institutions are given in Table 1 determined by the number of publications. The United States has the most publications and citations (193 articles, 7,994 citations), followed by China (186 articles, 1,555 citations), and Japan (131 articles, 2,034 citations), which account for nearly one-half the total number of publications in the field. Figure 3 depicts how a country cooperation network is used with VOSviewer and Scimago Graphica. The thickness and hue of the collaboration network connecting lines can show the degree of cooperation between countries or areas, with the closer the node color to red suggesting the intensity of the research collaboration. The analysis results indicate that the UK has the strongest international collaboration strength (with 68 total connection strengths), forming an extensive research collaboration network between the UK, the US, China, and Canada. The h-index is an estimate of a scholar's or country's scientific impact (18), and it balances the relationship between the number of published papers and their quality (the impact of the papers or the number of times they are cited) to provide a more complete picture of the impact of a scholar's research results. An examination of the PSD science field's h-index (Table 1) reveals that the United States has the greatest h-index (40) and total scientific research impact. The analysis of the PSD science field's h-index (Table 1) reveals that the United States has the greatest h-index (40) and total scientific research impact. However, it is worth mentioning that, despite accounting for a small number of publications, the United Kingdom and Canada rank second only to the United States in terms of article citations and h-index, and their significance should not be overlooked. Additionally, while China ranked second in terms of publications, it ranked in the middle of the pack in terms of h-index (23) and article citations (1555), indicating that the academic impact of the field is limited and that high-quality research needs to be conducted to increase the academic impact of the field.

3.3 Institutional analysis

The analyzed search results in WoSCC show that N8 Research Partnership is the institution with the highest number of publications and citations (46 articles, 2,322 citations), followed by the University of Manchester (36 articles, 1,913 citations), The State University System of Florida (32 articles, 2,160 citations), and the University of Florida (26 articles, 2098 citations), most of these institutions originated from Europe and the United States, showing a wide range of research interests and a strong influence in the field. VOSviewer has been utilized to create a network of institutional collaborations (Figure 4), with 114 institutions identified as having published at least five articles, and seven color clusters formed based on the intensity of collaboration between institutions, indicating a broader research collaboration between the clusters, with the purple and green sections each containing 23 institutions.

TABLE 1 Ranking of top 10 countries and institutions involved in the PSD rehabilitation field.

Rank	Country/region	Count (%)	Citations	H-index	Institution	Count (%)	Citations
1	USA	193 (17.78%)	7,994	40	N8 Research Partnership	46 (4.014%)	2,322
2	CHINA	186 (16.94%)	1,555	23	University of Manchester	36 (3.141%)	1913
3	JAPAN	131 (11.93%)	2034	24	The State University System of Florida	32 (2.79%)	2,160
4	SOUTH KOREA	97 (8.83%)	1,378	19	University of Florida	26 (2.27%)	2098
5	UNITED KINGDOM	92 (8.38%)	4,336	32	University of London	26 (2.27%)	1926
6	GERMANY	58 (5.28%)	1,444	21	Kumamoto Rehabillitation Hospital	23 (2.01%)	528
7	ITALY	56 (5.10%)	1,039	18	Seoul National University	21 (1.83%)	522
8	AUSTRALIA	49 (4.46%)	826	17	University College London	20 (1.75%)	1,276
9	CANADA	43 (3.92%)	3,545	25	University of Toronto	20 (1.75%)	2,727
10	SPAIN	39 (3.55%)	1,008	16	Autonomouw University of Barcelona	19 (1.66%)	1,276

10.3389/fneur.2023.1279452

FIGURE 3
Network of international cooperation of PSD rehabilitation in geographic visualization.

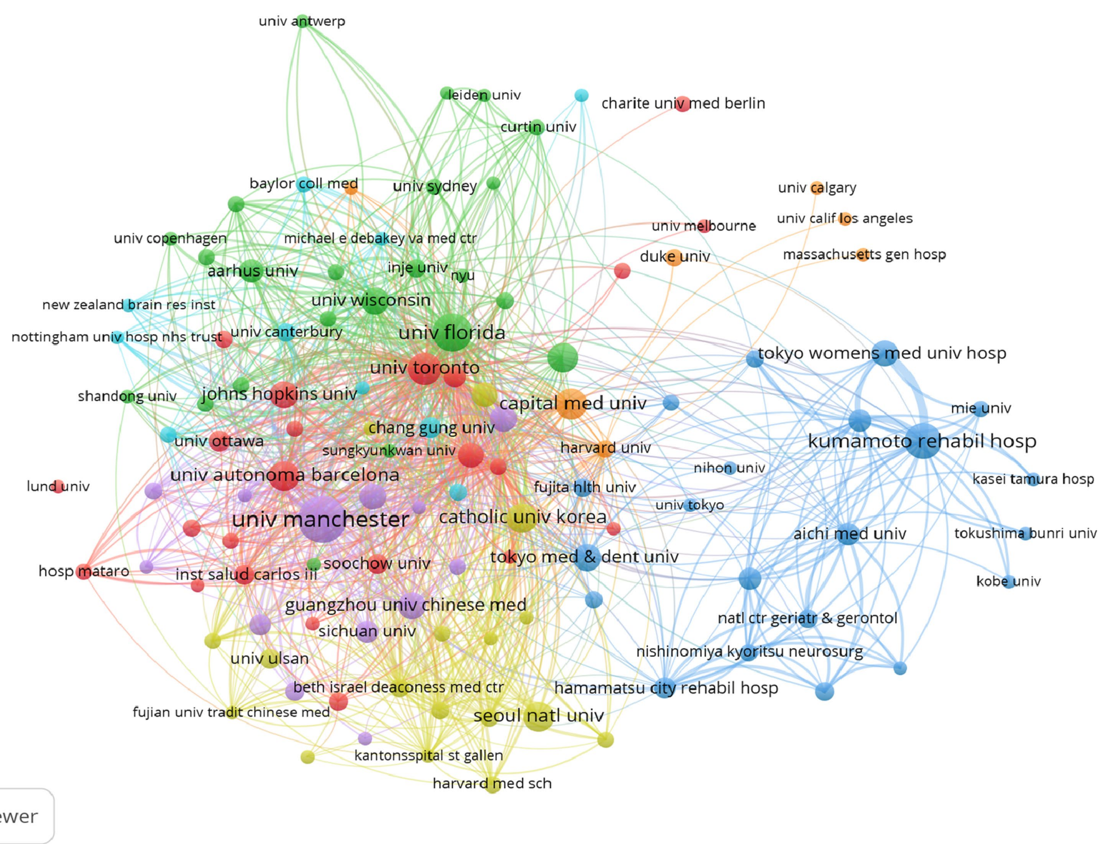


FIGURE 4
Institutional cooperation network diagram of PSD rehabilitation.

TABLE 2 Top 10 journals and co-cited journals of PSD rehabilitation.

Rank	Journal	Count(%)	IF (2022)	JCR	Co-cited journal	Citations	IF (2022)	JCR
1	Dysphagia	100 (8.73%)	2.6	Q2	Dysphagia	4,606	2.6	Q2
2	Archives of Physical Medicine and Rehabilitation	62 (5.41%)	4.3	Q1	Stroke	3,179	8.3	Q1
3	Journal of Stroke and Cerebrovascular Diseases	42 (3.67%)	2.5	Q3	Archives of Physical Medicine and Rehabilitation	1790	4.3	Q1
4	Frontiers in Neurology	22 (1.92%)	3.4	Q3	Journal of Stroke and Cerebrovascular Diseases	721	2.5	Q3
5	American Journal of Physical Medicine and Rehabilitation	20 (1.75%)	3.0	Q1	Neuron	660	16.2	Q1
6	Journal of Oral Rehabilitation	20 (1.75%)	2.9	Q2	Gastroenterology	492	29.4	Q1
7	Neurorehabilitation	19 (1.66%)	2.0	Q3	Journal of Neurologic Physical Therapy	479	3.8	Q2
8	Medicine	18 (1.57%)	1.6	Q4	Lancet	474	168.1	Q1
9	European Journal of Physical and Rehabilitation Medicine	17 (1.48%)	4.5	Q2	Clinical Neurophysiology	441	4.7	Q2
10	Stroke	17 (1.48%)	8.3	Q1	Cochrane Database of Systematic Reviews	985	8.4	Q1

3.4 Journal and co-cited journal distribution

Papers on PSD rehabilitation have been published in 374 scholarly publications since 1998. We used VOSviewer to perform a visual analysis of co-occurrence relationships between journals (Figure 5A) and co-citation relationships between co-cited journals (Figure 5B), and based on the results of the analysis, we plotted the top 10 journals and co-cited journals related to PSD rehabilitation (Table 2). Dysphagia took the top spot with 100 publications (8.73%), followed by the Archives of Physical Medicine and Rehabilitation (62 publications, 5.41%, 1,790 citations). Stroke (17 publications, 1.48%, 3,179 citations) was one of the top 10 journals with the highest impact. Stroke (17 articles, 1.1%, 3,179 citations) has the highest impact factor among the top 10 journals, as well as the most citations, demonstrating its strong academic importance in the subject of PSD rehabilitation. After analyzing the top 10 journals and co-cited journals, we discovered that six journals originated in the United States and four in the United Kingdom, which corresponds with the country/region and institutional contributions examined earlier and is sufficient to demonstrate the influence of these two countries in this research field.

Following this, we used CiteSpace to analyze the included literature with a dual map overlay (Figure 6), which links the cited journals on the left to the cited journals on the right to provide a clear, visual interpretation of citations for various combinations of publications, allowing for the identification of citation relationships, subject distributions, and patterns of movement across multiple disciplines (19). The ellipses on the map depict the number of publications relating to journals and the ratio of authors to the number of publications. The length of the ellipse represents the number of authors, the width of the ellipse represents the number of publications, and the curves between the left and right sides of the map are citation links, the trajectories of which provide an interpretation of the field's interdisciplinary relationships. The Z score thicker the links, the greater the score of the score function. Finally, we identified 5 major citation trajectories (pink and green), with publications in the fields of Neurology, Sports, and Ophthalmology (pink trajectory) influenced by publications in the domains of Molecular, Biology, and Genetics ($Z=3.46$, $f=2,192$), Health, Nursing, and Medicine ($Z=4.13$, $f=2,560$), and Psychology, Education, and Social ($Z=4.18$, $f=2,586$). Furthermore, past publications in the Health, Nursing, Medicine ($Z=2.79$, $f=2,822$) and Psychology, Education, and Social ($Z=2.11$, $f=1,442$) domains influenced publications in the Medicine, Medical, and Clinical (green track). Overall, this implies that the topic of PSD rehabilitation is strongly tied to basic scientific, clinical, nursing, and social subjects and that there is scope for multidisciplinary, collaborative study.

3.5 Authors and co-cited authors

To ensure the precision of data analysis, we initially leveraged the ResearcherID and ORCID functionalities of Web of Science to discern each author. Both systems bestow a distinct identifier upon every author, effectively addressing issues stemming from name ambiguities.

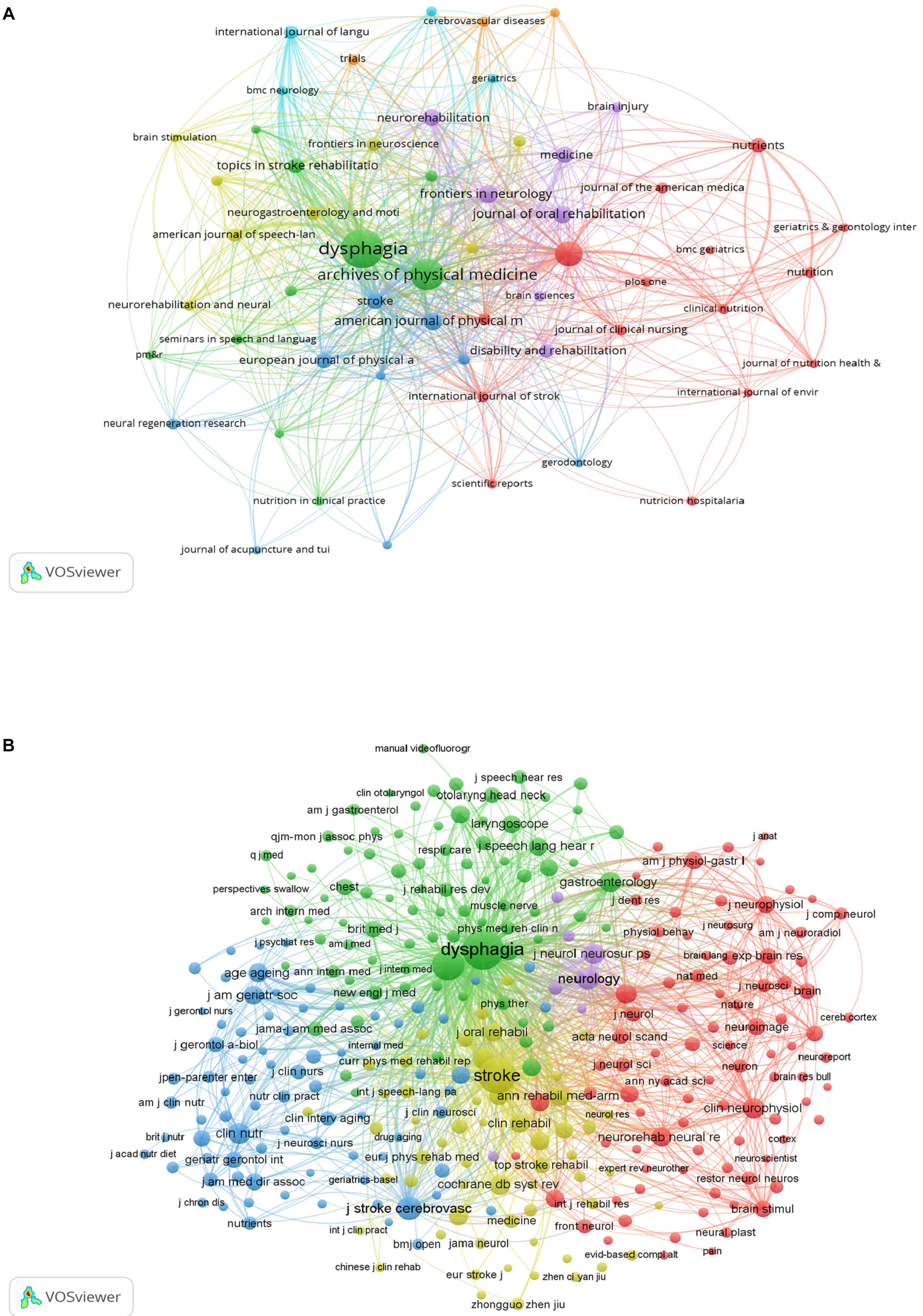


FIGURE 5
Analysis journals of PSD rehabilitation. **(A)** Journals co-occurrence analysis. **(B)** Co cited journals analysis.

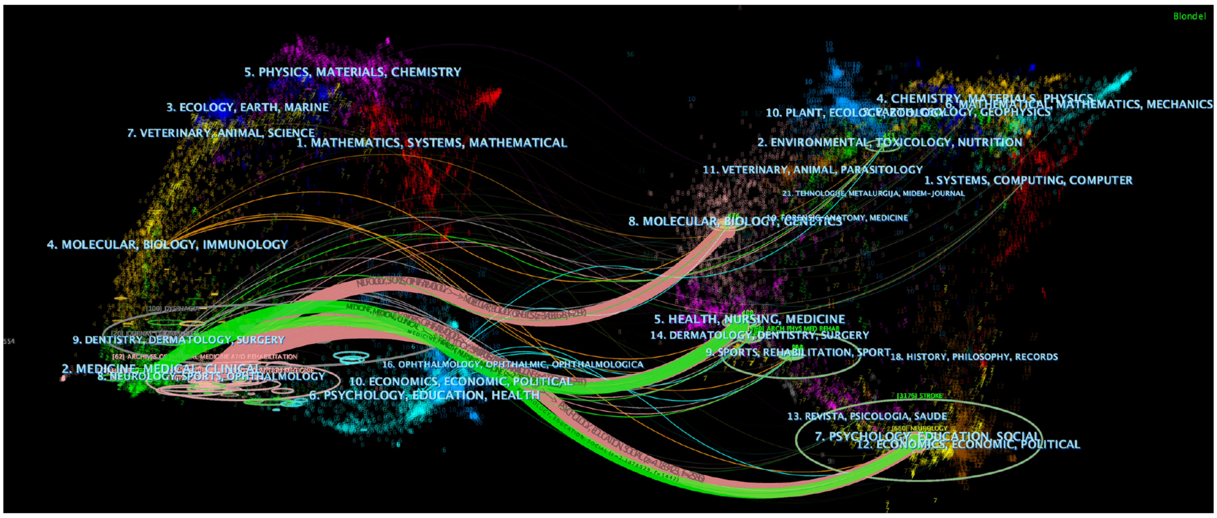


FIGURE 6
The dual map overlay of journals in PSD rehabilitation.

TABLE 3 Top 10 authors and co-cited authors of PSD rehabilitation.

Rank	Author	Count (%)	H-index	Co-cited author	Citations
1	Hamdy S	35 (3.00%)	20	Hamdy S	545
2	Wakabayashi H	26 (2.37%)	12	Martino R	461
3	Yoshimura Y	23 (2.09%)	11	Smithard DG	374
4	Nagano F	17 (1.55%)	8	Crary MA	348
5	Shiraishi A	16 (1.46%)	10	Logemann JA	342
6	Bise T	15 (1.37%)	8	Daniels SK	332
7	Shimazu S	15 (1.37%)	8	Mann G	285
8	Michou E	14 (1.27%)	11	Rosenbek JC	260
9	Momosaki R	13 (1.18%)	8	Robbins J	255
10	Park JS	13 (1.18%)	9	Langmore SE	199

Subsequently, before inputting data into the bibliometric analysis software, we inspected the raw data using Excel. Variations of names that unmistakably belonged to the same author were manually consolidated. Through our analysis, it was determined that the PSD rehabilitation domain encompassed contributions from a total of 4,991 authors. For our analysis, we used the Bibliometrix R package (4.1.3) in conjunction with R version 4.3.1, which allowed us to access representative scholars in the field to understand core research trends (20). The top 10 authors and co-citing authors of PSD rehabilitation research are shown in Table 3. The author with the most publications is Hamdy S from the University of Manchester (35 papers, 3.00%, h-index = 20), followed by Wakabayashi H (26 articles, 2.37%, h-index = 12) and Yoshimura Y (23 articles, 2.09%, h-index = 11). Hamdy S is also the most cited author (545 citations), with Martino R (461 citations) and Smithard DG (374 citations) coming in second and third, respectively. Figure 7A depicts a timeline of prominent authors in the field of PSD rehabilitation research, with the larger red circle representing the scholar's contribution to the discipline's research area. Hamdy S is a key pioneer in the subject, and his study "Recovery of swallowing after dysphagia stroke relates to functional reorganization in the intact motor cortex" is an example of his work (21). This paper investigates the mechanisms of swallowing recovery in PSD, implying a role for intact hemisphere reconfiguration in recovery and laying the groundwork for future research areas in PSD rehabilitation. The author cooperation network diagram (Figure 7B) depicts collaboration and relationships between authors and co-citing authors in the field. The author collaboration network formed by combining Figures 7A,B demonstrates that Martino R from the University of Toronto plays an important role in bridging the gap between research in the field, as demonstrated by his work "Dysphagia after stroke: incidence, diagnosis, and pulmonary complications, "which determined the prevalence of dysphagia and associated lung function impairment in stroke patients (2). The study has raised concerns among PTSD researchers about the link between dysphagia and pneumonia. Bath PM is a co-author of a new study in this field, "Swallowing therapy for dysphagia in acute and subacute stroke," which suggests that swallowing therapy may minimize hospitalization, dysphagia, and chest infections while also improving swallowing capacity. Chest infections, and may improve swallowing ability and provide new ideas for future swallowing therapy (22), providing new ideas for future

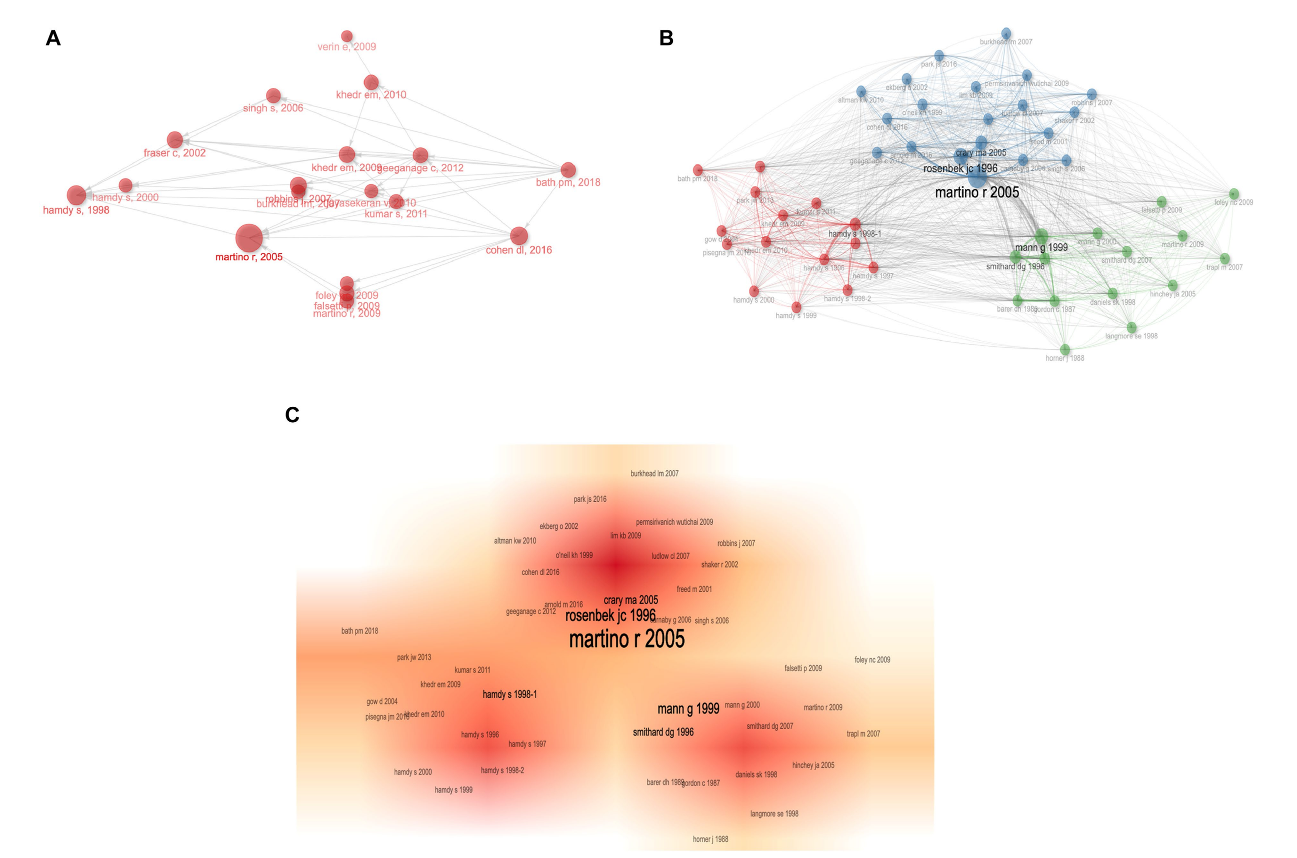


FIGURE 7
Co-authorship analysis of authors in PSD rehabilitation. **(A)** Historiography of influential author. **(B)** Network diagram of influential co-cited authors. **(C)** Influential co-cited author density visualization.

swallowing therapy. Figure 7C shows that the thicker and larger the author's name near the red colored block, the more influential that author is in the discipline, showing that Martino R is the most influential academic in this field of study.

3.6 Analysis of research hotspots

3.6.1 Publications with the highest number of citations

Table 4 shows the top 10 most referenced references in the PSD rehabilitation study domain. Among these, "Dysphagia after stroke: incidence, diagnosis, and pulmonary complications" had the most citations (326), and it was written by Martino R, which correlates to the previous study's findings. This matches the results of the preceding author's contribution analysis. In addition to being the most cited document, "Interventions for dysphagia and nutritional support in acute and subacute stroke" had the highest centrality (Centrality = 0.08). This article focuses on analyzing the impact of swallowing therapy, feeding, nutrition, and fluid supplementation on functional outcomes and death in patients with acute or subacute stroke with dysphagia (23), and it informs future research on nutritional support for stroke patients. After reviewing the 10 cited references, we discovered that this research focused on PSD etiology and neuroplasticity processes, intervention approaches, rehabilitation tactics, and complication prevention. CiteSpace was used to cluster the co-cited references (Figure 8A), CiteSpace provides two indicators, the module value (Q value) and the average profile value (S value), which can be used to judge the effectiveness of the mapping, the Q value is generally in the interval of [0, 1], the $Q > 0.3$ represents the division of the structure of the significant, the $S > 0.7$, that is, the clustering reaches a high efficiency, that is, the results are credible. The grouping coefficients are $Q = 0.8052$, and $S = 0.9207$. The color change from purple to yellow represents the time dimension, signifying a shift in study focus and trend. As seen in the picture, the field has progressed from a focus on pneumonia to investigating transcranial magnetic stimulation for the rehabilitation of PSD and is now concentrating on the prognosis of PSD, intervention dose, and dysphagia treatment.

3.6.2 Analysis of reference citation bursts

Using CiteSpace to identify the references with the strongest citation bursts can be used to anticipate future research frontiers. Figure 8B depicts the top 25 references that elicited the most powerful bursts, with the duration of the burst shown in red. "Post-stroke dysphagia: A review and design considerations for future trials" was the literature with the highest burst citation strength (strength = 16.33), and it focused on PSD pathogenesis, diagnosis, dysphagia management, pharmacologic treatment, and post-stroke pneumonia prevention from 2017 to 2021. Moreover,

TABLE 4 Top 10 co-cited references of PSD rehabilitation.

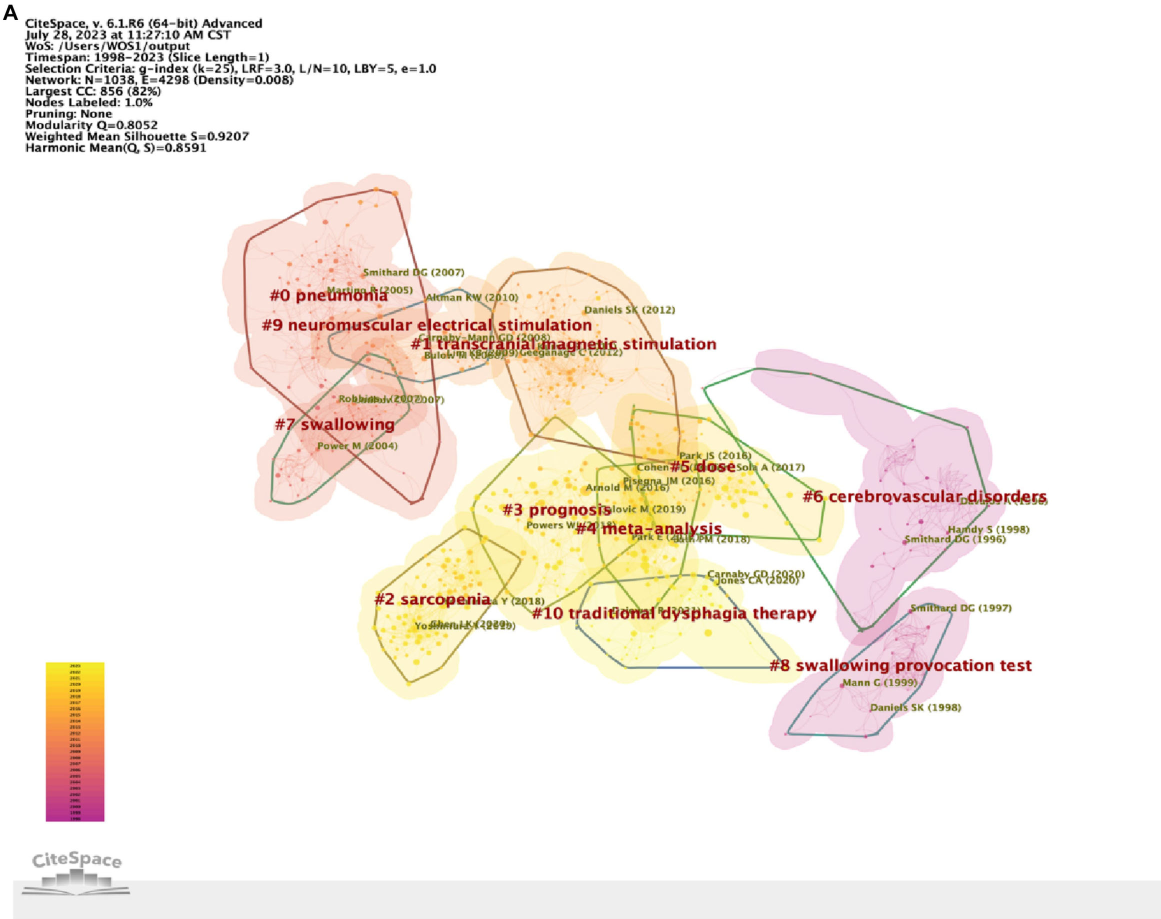
Rank	References	Cited frequency	Centrality	Document title	Source	IF (2022)	JCR
1	Martino et al. (2)	326	0.02	Dysphagia after stroke: incidence, diagnosis, and pulmonary complications.	Stroke	8.3	Q1
2	Crary et al. (24)	189	0.03	Initial psychometric assessment of a functional oral intake scale for dysphagia in stroke patients.	Archives of Physical Medicine and Rehabilitation	4.3	Q1
3	Hamdy et al. (21)	107	0.01	Recovery of swallowing after dysphagic stroke relates to functional reorganization in the intact motor cortex.	Gastroenterology	29.4	Q1
4	Cohen et al. (25)	84	0.06	Post-stroke dysphagia: A review and design considerations for future trials.	International Journal of Stroke	6.7	Q1
5	Robbins et al. (26)	70	0.06	The effects of lingual exercise in stroke patients with dysphagia.	Archives of Physical Medicine and Rehabilitation	4.3	Q1
6	Fraser et al. (27)	66	0.02	Driving plasticity in human adult motor cortex is associated with improved motor function after brain injury.	Neuron	16.2	Q1
7	Khedr et al. (28)	65	0.04	Treatment of post-stroke dysphagia with repetitive transcranial magnetic stimulation.	Acta Neurologica Scandinavica	3.5	Q2
8	Geeganage et al. (23)	57	0.08	Interventions for dysphagia and nutritional support in acute and subacute stroke.	Cochrane Database of Systematic Reviews	8.4	Q1
9	Falsetti et al. (29)	55	0	Oropharyngeal dysphagia after stroke: incidence, diagnosis, and clinical predictors in patients admitted to a neurorehabilitation unit.	Journal of Stroke and Cerebrovascular Diseases	2.5	Q3
10	Bath et al. (22)	54	0.01	Swallowing therapy for dysphagia in acute and subacute stroke.	Cochrane Database of Systematic Reviews	8.4	Q1

we discovered 5 co-cited references in the recent outbreak phase (22, 30–33). These 5 co-cited references in the recent outbreak are primarily concerned with the topics of pathogenesis and neuroplasticity mechanisms of PSD, swallowing treatment, and rehabilitation effects.

3.6.3 Clustering analysis and keyword occurrence frequency

The term cluster analysis can summarize research topics and aid in understanding research hotspots in the subject. VOSviewer estimated 3,250 keywords, and the terms with a frequency of

A



B

Top 25 References with the Strongest Citation Bursts

References	Year	Strength	Begin	End	1998 - 2023
Martino R, 2005, STROKE, V36, P2756, DOI 10.1161/01.STR.0000190056.76543.eb, DOI	2005	11.04	2008	2010	
Carnaby G, 2006, LANCET NEUROL, V5, P31, DOI 10.1016/S1474-4422(05)70252-0, DOI	2006	7.17	2008	2011	
Smithard DG, 2007, AGE AGEING, V36, P90, DOI 10.1093/ageing/afl149, DOI	2007	7.52	2009	2012	
Bulow M, 2008, DYSPHAGIA, V23, P302, DOI 10.1007/s00455-007-9145-9, DOI	2008	10.31	2010	2013	
Lim KB, 2009, J REHABIL MED, V41, P174, DOI 10.2340/16501977-0317, DOI	2009	6.93	2010	2014	
Kumar S, 2011, STROKE, V42, P1035, DOI 10.1161/STROKEAHA.110.602128, DOI	2011	7.7	2012	2016	
Geeganage C, 2012, COCHRANE DB SYST REV, V0, P0, DOI 10.1002/14651858.CD000323.pub2, DOI	2012	10.7	2013	2017	
Daniels SK, 2012, STROKE, V43, P892, DOI 10.1161/STROKEAHA.111.640946, DOI	2012	7.63	2013	2017	
Michou E, 2012, GASTROENTEROLOGY, V142, P29, DOI 10.1053/j.gastro.2011.09.040, DOI	2012	7.11	2013	2017	
Michou E, 2014, BRAIN STIMUL, V7, P66, DOI 10.1016/j.brs.2013.09.005, DOI	2014	6.99	2015	2018	
Pisegna JM, 2016, CLIN NEUROPHYSIOL, V127, P956, DOI 10.1016/j.clinph.2015.04.069, DOI	2016	9.73	2016	2021	
Lim KB, 2014, ANN REHABIL MED-ARM, V38, P592, DOI 10.5535/arm.2014.38.5.592, DOI	2014	8.94	2016	2019	
Cohen DL, 2016, INT J STROKE, V11, P399, DOI 10.1177/1747493016639057, DOI	2016	16.33	2017	2021	
Park JS, 2016, J ORAL REHABIL, V43, P426, DOI 10.1111/joor.12390, DOI	2016	8.43	2017	2021	
Suntrup S, 2015, EUR J NEUROL, V22, P832, DOI 10.1111/ene.12670, DOI	2015	8.09	2017	2020	
Nii M, 2016, J STROKE CEREBROVASC, V25, P57, DOI 10.1016/j.jstrokecerebrovasdis.2015.08.033, DOI	2016	7.39	2017	2019	
Arnold M, 2016, PLOS ONE, V11, P0, DOI 10.1371/journal.pone.0148424, DOI	2016	13.99	2018	2021	
Chen YW, 2016, CLIN REHABIL, V30, P24, DOI 10.1177/0269215515571681, DOI	2016	8.38	2018	2021	
Bath PM, 2016, STROKE, V47, P1562, DOI 10.1161/STROKEAHA.115.012455, DOI	2016	7.65	2018	2021	
Park JS, 2016, J ORAL REHABIL, V43, P364, DOI 10.1111/joor.12382, DOI	2016	7.28	2018	2021	
Park E, 2017, BRAIN STIMUL, V10, P75, DOI 10.1016/j.brs.2016.08.005, DOI	2017	8.34	2020	2023	
Yoshimura Y, 2019, NUTRITION, V61, P111, DOI 10.1016/j.nut.2018.11.005, DOI	2019	8.06	2020	2023	
Galovic M, 2019, JAMA NEUROL, V76, P561, DOI 10.1001/jamaneurol.2018.4858, DOI	2019	7.22	2020	2023	
Bath PM, 2018, COCHRANE DB SYST REV, V0, P0, DOI 10.1002/14651858.CD000323.pub3, DOI	2018	10.74	2021	2023	
Chen LK, 2020, J AM MED DIR ASSOC, V21, P300, DOI 10.1016/j.jamda.2019.12.012, DOI	2020	9.7	2021	2023	

FIGURE 8
References analysis of PSD rehabilitation. **(A)** Co-citation references clustering. **(B)** Top 25 references with the strongest citation bursts.

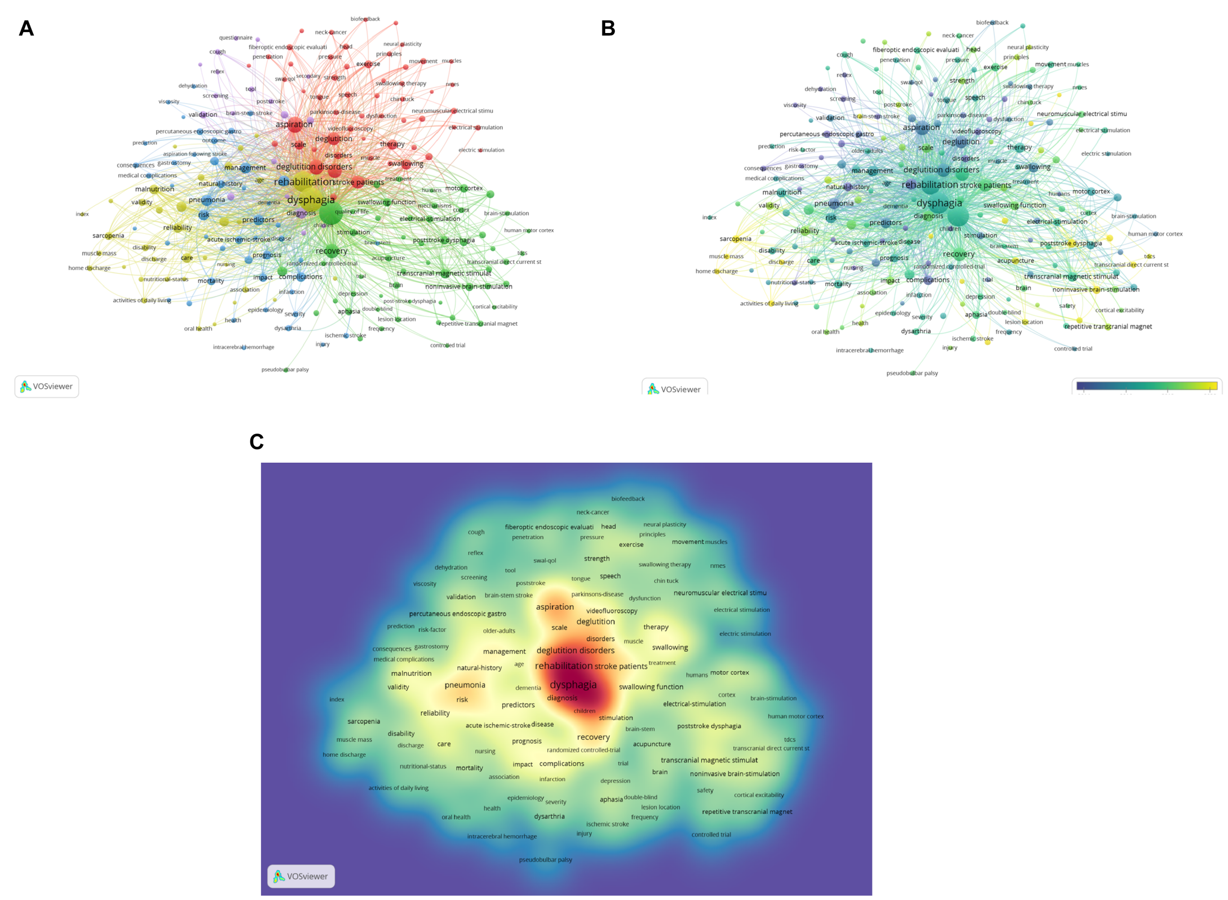


FIGURE 9
Keywords analysis of PSD rehabilitation. **(A)** Cluster view of keywords. **(B)** Time-overlapping visualization of keywords. **(C)** The density visualization map of keywords.

occurrence higher than 5 times or more were removed and incorporated in the co-occurrence analysis, resulting in 5 color clusters (Figure 9A), which indicate 5 study directions. In addition to "dysphagia" and "rehabilitation," the main keywords in the yellow clusters also included "malnutrition ", "reliability," "validity," and "care"; the main keywords in the green cluster are "quality of life," "recovery," "stimulation," "transcranial magnetic stimulation" and "noninvasive brain-stimulation"; in the red cluster, the main keywords were "deglutition disorders," "aspiration," "deglutition," "scale," "therapy"; the main keywords in the blue cluster are "management," "pneumonia," "predictors," "risk"; the main keywords in the purple cluster are "diagnosis ", "validation," "tool," "brain-stem stroke." Except for "dysphagia," "rehabilitation," and "deglutition disorders," the terms "diagnosis," "pneumonia," "risk," and "aspiration" had the most weight, according to the keyword density map (Figure 9B). We also created a temporal overlap representation of the keywords (Figure 9C), in which the gradient from purple to yellow represents the period of the keywords, with yellow signifying current occurrences of the keywords, which represent recent periods of study in the subject.

3.6.4 Keyword citation burst analysis and trend topics

Burst keywords are high-frequency keywords that erupt at a specific moment, showing the emergence of hotspots in the study area and anticipating research trends. Figure 10A depicts the top 25 terms with the highest number of citations, with "natural history," "percutaneous endoscopic gastrostomy," and "predictor" being the top three keywords with the most citation numbers. Furthermore, the terms that received the most attention were "consequence" (2000–2012), "brain stem stroke" (2000–2014), "acute stroke" (2002–2015), and "complication" (2001–2011). Excluding words related to the search term, "validity," "noninvasive brain-stimulation," "home discharge," "muscle mass," "guideline," and "sarcopenia," "rTMS" are the keywords of citation explosion during 2020–2023, which are regarded as the hotspots that researchers pay high attention to shortly, among which "validity" and "noninvasive brain-stimulation" are the keywords. Among these, "validity" and "non-invasive brain stimulation" are the most powerful. We also used Bibliometrix R to look for trending themes in the field of PSD rehabilitation (Figure 10B). Keywords like "health care professionals," "transcranial direct current stimulation (tDCS)," and "acupuncture" are suggestive

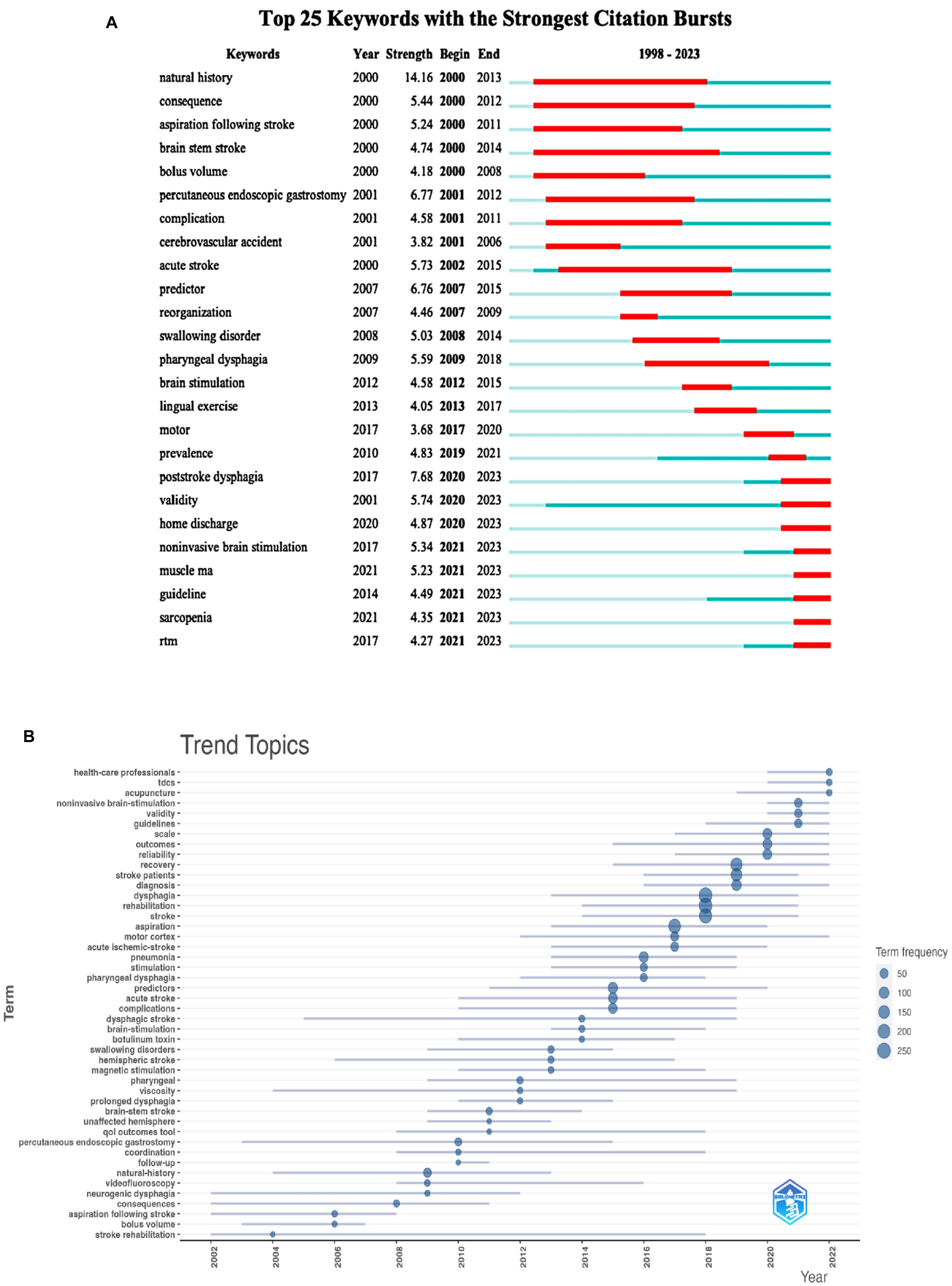


FIGURE 10
Keywords analysis of PSD rehabilitation. **(A)** Top 25 keywords with the strongest citation bursts. **(B)** Trend topics of keywords.

of future research trends. Overall, research hotspots in the field of PSD rehabilitation focus on PSD-related guidelines, the accuracy, and validity of screening tools, noninvasive brain stimulation, transcranial magnetic stimulation (rTMS), tDCS, and acupuncture for PSD rehabilitation.

4 Discussion

4.1 General information

In this study, we used CreateSpace, VOSviewer, and Bibliometrix R to assess the literature on PSD rehabilitation. The results of the analysis show that the trend of growth in the number of annual publications over the last 25 years has been divided into two phases, with slow growth in the number of publications between 1998 and 2013, a period that may be related to the increased focus of research in the field on the incidence of stroke and the treatment of the disease. The number of publications begins to increase between 2014 and 2023, with the most pronounced trend of growth in 2022, which is likely to be explained by Guidelines issued by the European Stroke Organization and the European Swallowing Society on the diagnosis and treatment of post-stroke dysphagia (34). The guideline proposes the impact of PSD on stroke outcomes, nutritional screening for dysphagia, dysphagia assessment, and PSD treatment, and it provides evidence-based recommendations for the management of PSD by a multidisciplinary team aimed at preventing, diagnosing, and treating PSD, as well as bringing PSD rehabilitation to the attention of a broader range of researchers and providing ideas for future studies.

The examination of national and institutional collaboration networks reveals that research in the field of PSD rehabilitation dominates in industrialized nations. On the one hand, this may be attributable to the significance that industrialized nations have on PSD rehabilitation, and on the other, it may be connected to the level of the national economy, policy, and financial assistance. The United States has not just the most publications, but also the most citations. The United Kingdom does not have the most publications, but it is second only to the United States in terms of citations and the h-index, and it is one of the top countries in terms of total institutional effect and worldwide influence. China has the most common stroke cases in the world (35), and as a result, it places a high priority on stroke-related research, which explains why China comes second in terms of the number of publications. However, while China has a high number of publications, its citation, and h-index rankings are moderate, indicating that the academic impact has not been matched and that, in the future, it should focus on innovation and high-quality research in the subject area to improve the quality of articles.

According to the findings of this investigation, Dysphagia is the journal with the most articles and citations. Except for three journals that are Q1, the majority of the top 10 are Q2 or Q3, with an average IF of 3.51, showing that the field's academic influence still needs to be enhanced. According to the findings of the study, Hamdy S was not only the author with the most publications, but also the author with the highest co-citation rating. The primary emphasis of this author's study was on the mechanics of swallowing recovery in PSD (21), which provided a solid foundation for further research on PSD rehabilitation. The second most prominent author in the area is Martino R., whose work first revealed the association between PSD and the prevalence of pneumonia (2), promoting researchers in the field to re-examine PSD and perform in-depth research on PSD and pneumonia.

The studied search results in WoSCC revealed that papers on PSD rehabilitation include a wide range of topic areas, with the top three being Rehabilitation (259 articles), Clinical Neurology (225 articles), and Neurosciences (206 articles). Despite the limited number of papers (78), Peripheral Vascular Disease has the largest amount of single citations (58.95). In addition, the papers include Otorhinolaryngology, General Internal Medicine, Peripheral Vascular Disease, Sports Sciences, Nutrition Dietetics, and Audiology Speech Language Pathology. Furthermore, a biplot overlay analysis of published journals reveals that PSD rehabilitation is influenced by basic scientific, clinical, nursing, and social subjects, implying that PSD rehabilitation is a complex area of research requiring multidisciplinary collaboration and intervention implementation.

4.2 Research hotspots and trends

First, reference co-citations and keyword co-occurrences are investigated in this study to discover research topics and trends in the field of PSD rehabilitation. Second, the findings of temporal overlap visualization and keyword burst detection were utilized to highlight research hotspots and frontiers in the field. Finally, the field's hotspots and cutting-edge developments were highlighted, covering the pathophysiology and neuroplasticity processes of PSD, comorbidities, swallowing screening and evaluation approaches, and swallowing rehabilitation modalities. We will examine the aforementioned hotspots and cutting-edge developments to give references and lessons for researchers in the field of PSD rehabilitation.

4.2.1 Pathogenesis of PSD and mechanisms of neuroplasticity

According to data, the incidence of dysphagia in hemorrhagic stroke is between 58.6 and 67% (36, 37). The cortex is important in swallowing control, and subcortical lesion sites are usually associated with swallowing dysfunction. Even a small volume of hemorrhage can disrupt the neural network of swallowing, leading to swallowing difficulties, which is why hemorrhagic stroke patients have a high incidence of dysphagia (38). Dysphagia occurs in 32.3% of ischemic stroke patients, with subcortical infarction being more likely to develop moderate to severe dysphagia, and individuals with dysphagia generally have bilateral vertebral tract injury (39–41). According to research, bilateral subcortical lesions are major determinants impacting patient prognosis (42), and physicians must pay attention to this prognostic indicator to assess the patient's prognosis. Furthermore, several studies have shown that patients with PSD have a poor neurophysiological response in the right hemisphere, dysphagia caused by infratentorial stroke is more severe than dysphagia caused by supratentorial stroke, and the right lenticular nucleus is associated with the development and severity of dysphagia (43–45).

According to multicenter retrospective cohort research, the majority of stroke patients have sarcopenia, and there is an independent negative link between sarcopenia and the recovery of swallowing function (46). Another research validated the link between temporal muscle thickness and dysphagia in stroke patients, demonstrating that temporal muscle thickness is an independent risk factor for dysphagia in patients with acute stroke (47). It is essential to note that significant muscular weakness and muscle volume loss can cause delayed dysphagia longer than 7 days after a stroke, therefore it is critical to recognize and diagnose early and execute early management (48). The foregoing findings open up new avenues for future study, and it is critical to assess if stroke patients have dysphagia caused by muscle loss to give early and personalized preventative and therapeutic approaches.

The brain is capable of neuroplasticity, and the cerebral cortex has an evident hemisphere response to swallowing. It has been discovered that individuals with recurrent cerebral infarction and central dysphagia exhibit compensatory remodeling of neurological function (49). Functional magnetic resonance imaging (fMRI) is a useful tool for studying the neurophysiology of swallowing *in vivo*, and it has been used to study the manifestations of the neural control of swallowing in normal subjects and patients with dysphagia, as well as the effects of swallowing therapy on neuroplasticity (50). Stroke is classified into four phases: hyperacute, acute, subacute, and chronic, with the window of greatest neural plasticity in the subacute phase also being a sensitive period for stroke recovery, and the potential for inducing recovery in the chronic phase becoming limited over time (51). Even though the chronic phase of stroke often lacks neuro independent recovery (52, 53), recovery is still achievable with intense neurorehabilitation, but at a significantly lower pace than in the subacute period (54). Functional connectivity between the cortex and the medulla was found to be enhanced in patients with episodic stroke, and functional connectivity between the cortex and the medulla could serve as a biomarker of swallowing-related changes in the neural network of the brain, implying that the brain's precentral gyrus plays an important role in the regulation of neuroplasticity in pharyngeal swallowing (55). Increased excitability of the swallowing cortical bulb is primarily located in the undamaged cerebral hemisphere, and the sensory-driven human motor cortex is largely dependent on the frequency, intensity, and duration applied to promote neuroplasticity after brain injury (23), suggesting that there may be individual differences in the rehabilitation effects of PSD patients after using the same swallowing function rehabilitation intervention method. As a result, it is critical to analyze the patients' degree of swallowing impairment and select the optimal intervention method to accomplish the therapeutic impact in the future rehabilitation process.

4.2.2 Complications of PSD

Patients with PSD have a 4.35 times higher prevalence of pneumonia and are up to 4.07 times more likely to die than non-PSD patients, and the risk of PSD is strongly associated with hemorrhagic strokes, a history of previous strokes, severe strokes, gender (women are more likely to have a stroke than men), and diabetes mellitus (56). The severity of PSD modifies the patient's food patterns, and alterations in eating pathways increase the likelihood of respiratory infections (57). According to studies, more than a quarter of patients with subarachnoid hemorrhage are not evaluated for dysphagia, and one-fifth of these patients acquire pneumonia (58) and delays in dysphagia screening and evaluation increase the risk of pneumonia infection. As a consequence, detecting and treating dysphagia in stroke patients at an early stage can help minimize the risk of pneumonia and death (2, 59). Training the respiratory muscles after a stroke can prevent infiltration or aspiration when swallowing fluid push, lowering the risk of respiratory problems and improving dysphagia (60). Malnutrition and insufficient fluid intake are common in PSD patients (61, 62), and malnutrition hurts stroke patients' recovery of physical and swallowing function, prolonging hospitalization, increasing the burden of care, and increasing mortality. Nutritional screening and evaluation can help predict swallowing and physical function results in patients. As a result, there is a need for early nutritional evaluation and the adoption of early nutritional therapies that promote swallowing function recovery and enhance patient prognosis (63, 64). There is a risk of dehydration for patients after an acute stroke, particularly those with PSD, and optimal fluid replacement appears to be extremely important in reducing the risk of neurological deterioration and other complications; however, there is a lack of a clear method of hydration status assessment, a shortage of time for healthcare staff, unclear work patterns, and a low emphasis on hydration status management (65). Healthcare collaboration is critical in the treatment of hydration status after stroke, and there is a need to better define the stages and techniques of hydration status evaluation, as well as to raise healthcare personnel's awareness of hydration status management to satisfy patients' rehydration needs.

4.2.3 Swallowing screening methods

Swallowing screening is one of the hottest subjects in the world of PSD rehabilitation research. Guidelines recommend that stroke patients be screened for swallowing before oral intake (such as medications, food, and liquids) and that abnormalities on initial screening be referred to a clinician trained in Speech-Language Pathology, Occupational Therapy, Dietitian, or Trained Dysphagia Clinician for a more detailed bedside para pharyngeal swallowing assessment and management of swallowing, feeding, nutrition, and hydration status (66). Among the swallowing screening evaluation procedures, the Water Swallowing Test (WST) and the Gugging Swallowing Screen (GUSS) are the most widely used and actionable. The Water Swallowing Test can determine whether a patient has significant malabsorption and whether instrumentation is required to further assess dysphagia to reduce unnecessary instrumentation; however, it is difficult to identify patients with silent malabsorption and carries the risk of false negatives (67). The GUSS is primarily indicated for acute stroke patients; the assessment method takes into account the case physiology of voluntary swallowing, and allows for the use of foods of varying consistencies in the swallow test to reduce the risk of aspiration in acute stroke patients due to difficulty swallowing liquids, has a high sensitivity (close to 100%), and is a reliable tool for the detection of dysphagia and the screening of patients at high risk of aspiration (68, 69). The Volume-viscosity swallow test (V-VST) is used to examine dysphagia using fluids and thickeners in conjunction with pulse oximetry. It may be used not only for screening but also to offer reliable indications of ideal fluid push volume and viscosity as well as therapy suggestions. V-VST, on the other hand, is reliant on the prevalence of the context or clinical situation in which it is used, with a positive predictive value of 68.3% (false positives as high as 31.7%) when the prevalence does not reach 30, and 95.2% in populations with a prevalence of 80% (70). Videofluoroscopy (VFS) and fiberoptic endoscopic evaluation (FFE) of swallowing are more accurate instrumental evaluations for assessing swallowing function and providing information on swallowing physiology, anatomy, and function. However, they have significant limitations in their use, as they are susceptible to patient posture, somatic status, cognition, and acceptance, secondly, the operation of VFS and FFE requires specialized equipment and training, and it is currently not possible to identify a swallowing screening tool that has high accuracy, sensitivity, specificity, and is perfect (8, 71), and a combination of approaches may be required.

4.2.4 Rehabilitation for PSD

The emphasis of the keyword analysis study is neurorehabilitation. The most explored rehabilitative methods with the highest research enthusiasm are repeated transcranial magnetic stimulation (rTMS), transcranial direct current stimulation (tDCS), and acupuncture. rTMS is a non-invasive brain stimulation technique that reverses the pharyngeal motor cortex, modulates brain activity and swallowing motor behavior, and also modulates neurotransmitters, activates/polarizes immune cells (astrocytes and microglia), and inflammatory cytokines in the brain, thereby affecting brain function and improving post-stroke dysfunction (72, 73). A meta-analysis has shown that rTMS not only enhances swallowing function and daily life activities for PSD patients and reduces the incidence of aspiration but also proves to be a safe and feasible rehabilitation method (74), however, to accomplish excellent recovery, attention should be required to determine the proper stimulation intensity during the intervention. tDCS can improve dysphagia recovery after stroke by promoting reconfiguration of the swallowing neural network, so expediting dysphagia recovery after acute stroke, and the earlier therapy is started, the better the outcome of dysphagia recovery (75). A meta-analysis, however, found that tDCS was helpful for dysphagia after unilateral hemisphere stroke, medullary palsy, and brainstem stroke, but not for ataxia and dysphagia after basal ganglia stroke (76). Furthermore, because tDCS may be susceptible to the baseline level of the study population, such as age, affected cerebral hemispheres, comorbidities, and complications after acute stroke, attention should be paid to the program design of tDCS in rehabilitation. As a result, in the case of tDCS interventions, it is common to combine tDCS with motor-specific or peripheral sensory-motor stimulation and to combine cerebral stimulation with sensory feedback to entrainment (77). Furthermore, in terms of neurostimulation therapies such as neuromuscular electrical stimulation (NMES), pharyngeal electrical stimulation (PES), and transcranial direct current stimulation (tDCS), a combined approach would be more effective than single neurostimulation therapies or traditional dysphagia therapies in improving PSD (78). Acupuncture affects the bilateral cerebral hemispheres via feature brain areas of interest (ROIs), reduces aberrant functional connectivity, and enhances motor cortex recovery after stroke. By combining intervention with swallowing training, acupuncture can significantly boost the rehabilitative result (79, 80). Although evidence for the potential therapeutic alternatives and efficacy of acupuncture continues to emerge, international standards to assist clinical practice and treatment have yet to be produced (81).

In addition to these three hot rehabilitation modalities, intermittent theta burst stimulation (iTBS) is a non-invasive brain stimulation technique. By bidirectionally enhancing the excitability of the motor cortex associated with the genioglossus muscle and promoting neural remodeling, it effectively augments the swallowing function in patients with post-stroke dysphagia (82). It's worth noting that, compared to rTMS, the combination of iTBS with swallowing exercises demonstrates a more pronounced improvement in the excitability of the swallowing motor cortex and swallowing function following a stroke. When combined with swallowing exercises that stimulate both cerebellar hemispheres, iTBS significantly reduces scores on the Fiberoptic Endoscopic Dysphagia Severity Scale (FEDSS), the Penetration-Aspiration Scale (PAS), and the Standard Swallowing Assessment (SSA). Concurrently, it notably elevates scores on the Functional Oral Intake Scale (FOIS), effectively improving swallowing function (83). Transcutaneous auricular electrical vagus nerve stimulation (ta-VNS) modulates swallowing function via bilateral extra-auricular stimulation of the vagus nerve, with few serious adverse events occurring during the intervention, which is safe and can be used as a novel non-invasive treatment strategy (84). Furthermore, the involvement of speech-language pathologists (SLPs) and nurses in the management of PSD rehabilitation is critical, and this co-management model can assist the rehabilitation team in consistently obtaining safety information about dysphagia and facilitating the implementation of the rehabilitation program; however, implementable options are needed to be explored further (85).

5 Strengths and limitations

The bibliometric analysis demonstrates significant differences and advantages over other online tools in the realm of academic research. Firstly, bibliometric analysis is specifically tailored to peer-reviewed scholarly articles, ensuring depth, breadth, and high quality of data. This level of specificity allows researchers to gain a comprehensive understanding of a research field, a depth that many online tools often fall short of. Additionally, it offers analyses spanning extended timeframes, making it possible to reveal evolving research trends, a feature not easily found in other tools. Using tools such as CiteSpace and VOSviewer for bibliometric studies enables an in-depth exploration of relationships among research topics and provides a structured overview. Concurrently, it relies on standardized academic databases, ensuring the data's scientific integrity, accuracy, and consistency. This mode of analysis also offers a quantitative approach for researchers to evaluate the impact and significance of their work and aids in unveiling patterns and trends in research collaborations. While some online tools may boast advantages in real-time updates, visualization, or user interface, bibliometric analysis indisputably excels in depth, scope, and scientific rigor. Therefore, to provide a more comprehensive and systematic insight, we employed the bibliometric analysis approach to study literature related to post-stroke dysphagia rehabilitation, culminating in a dedicated research article.

Using the CiteSpace, VOSviewer, and Bibliometrix R software packages for bibliometric analysis, this study synthesizes the strengths of the three software packages to provide an overview of the research progress and cutting-edge trends in the field of PSD rehabilitation around the world, allowing researchers to quickly understand the current state of research and hotspots in the field. However, because this study is not a substitute for a systematic review, there are certain limitations to this review. First, due to the limitations of the software and research methodology, we only selected literature from the WoSCC database, whereas most other databases, such as PubMed, Embase, and Scopus, do not have comprehensive information about their full text and citation analysis, which is why we chose the WoSCC database. As a result, this may neglect the contribution of literature from other databases in this study field. Second, we merely screened the retrieved literature based on the inclusion and exclusion criteria and did not analyze the quality of the literature, which might be biased. Third, certain high-quality publications may be ignored in the literature analysis process due to their recent publication and low citation counts, which may contribute to bias. However, the WoSCC database has a huge amount of high-quality core literature from all around the world, and we continue to feel that our study covers the research hotspots and frontiers of PSD rehabilitation and can give certain information for future research.

6 Conclusion

In this study, the WoSCC database was searched for the literature on PSD rehabilitation from the previous 25 years, and the titles and abstracts of the literature were read and screened according to the inclusion and exclusion criteria, and those that met the criteria requirements were included as required. The study of the number of publications, nations, institutions, authors, co-cited references, and keywords using the CiteSpace, VOSviewer, and Bibliometrix R software packages explains the general picture of PSD rehabilitation research and highlights research hotspots and cutting-edge trends. The visualization analysis findings reveal that research on this topic is gaining more and more attention from researchers, the number of publications is increasing, and the research field is in the process of continuous exploration. According to the study, future research hotspots and frontiers will mostly cover the pathophysiology and neuroplasticity processes of PSD, comorbidities, swallowing screening and evaluation methodologies, and swallowing rehabilitation modalities. These research goals and frontiers underscore the significance of PSD management in the swallowing rehabilitation process. Overall, this work may successfully give a research trajectory of PSD rehabilitation as well as significant research information and research suggestions to researchers interested in this subject.

Author contributions

YH: Data curation, Formal analysis, Software, Visualization, Writing – original draft. XT: Data curation, Formal analysis, Software, Visualization, Writing – original draft. HK: Writing – review & editing. HW: Data curation, Software, Writing – review & editing. YX: Writing – review & editing. DZ: Supervision, Writing – review & editing. CL: Supervision, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Research hotspots and frontiers in post-stroke dysphagia: a bibliometric analysis study

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Background: Dysphagia is a common complication of stroke that can result in serious consequences. In recent years, more and more papers on post-stroke dysphagia have been published in various journals. However, there is still a lack of bibliometric analysis of post-stroke dysphagia. This study visually analyzes the global research situation of post-stroke dysphagia from 2013 to 2022, aiming to explore the current research status, frontier trends, and research hotspots in this field.

Methods: Articles and reviews relevant to post-stroke dysphagia were obtained and retrieved from the Web of Science core collection database in the last 10 years (from 2013 to 2022). CiteSpace and Microsoft Excel 2019 were used for bibliographic analysis.

Results: A total of 1,447 articles were included in the analysis. The number of publications showed an overall upward trend, from 72 in 2013 to 262 in 2022. The most influential authors, institutions, journals, and countries were Hamdy S, University of London, Dysphagia, and the People's Republic of China. An analysis of keywords and the literature indicated that current studies in the field of post-stroke dysphagia focused on dysphagia and aspiration, dysphagia classification, dysphagia rehabilitation, and daily living.

Conclusion: This bibliometric analysis reveals the latest advancements and emerging trends in the field of post-stroke dysphagia, spanning the years 2013 to 2022. It highlights the paramount importance of conducting large-scale randomized controlled trials examining the efficacy of dysphagia screening protocols and non-invasive intervention techniques in improving the quality of life for these patients. Such research efforts hold significant academic implications for the development of evidence-based treatment strategies in this field.

KEYWORDS

dysphagia, stroke, CiteSpace, visual analysis, bibliometric

Introduction

Stroke is a group of cerebrovascular diseases caused by organic brain damage, with common clinical characteristics of sudden onset and rapid development of localized or diffuse brain function deficits (1, 2). Stroke is the main cause of disability and death (3). Due to the aging population, high blood pressure, and other risk factors, as well as poor management, the

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incidence rate of stroke continues to rise, which will further increase the emotional and economic burden on families and society (4–6). Swallowing is one of the most basic physiological activities that humans rely on for survival (7). Depending on the part of the food passing through, it can be generally divided into oral, pharyngeal, and esophageal stages. Any structural or functional damage in any of these stages can lead to the occurrence of dysphagia (8–10). Dysphagia is a common complication of stroke, and relevant data show that 28 to 67% of stroke patients have dysphagia (11).

The diagnosis of post-stroke dysphagia is relatively easy, but determining the location and nature of dysphagia requires detailed clinical evaluation and instrumental examination (12). Clinical evaluation includes dysphagia screening or a comprehensive evaluation of the orofacial structure and function by a speech and language pathologist. Instrumental evaluation includes the Video Fluoroscopic Swallowing Study (VFSS) and Flexible Endoscopic Evaluation of Swallowing (FEES). Post-stroke dysphagia can cause an inability to eat normally and can lead to serious consequences such as malnutrition, aspiration pneumonia, and psychological disorders, resulting in prolonged hospitalization and increased complications (13–15). Treatment for post-stroke dysphagia includes dietary interventions, behavioral interventions, nutritional interventions, interventions to improve oral health, pharmacological treatment, and neurostimulation treatment (16). For instance, research has shown that nutrients given through the gastrointestinal tract are more easily absorbed and help maintain the integrity of the intestinal mucosal structure and intestinal barrier. Therefore, enteral nutrition such as nasogastric tubes and percutaneous endoscopic gastrostomy (PEG) are of great significance for patients with post-stroke dysphagia (17). Although there is increasing consensus on the effectiveness of existing treatment methods in promoting the rehabilitation of post-stroke dysphagia, there are still many problems and uncertainties that need to be explored. Therefore, research on post-stroke dysphagia is necessary and valuable. In recent years, more and more papers on post-stroke dysphagia have been published in various journals (18–21). However, there is still a lack of scientific and measurement analysis of post-stroke dysphagia.

Bibliometric analysis uses quantitative methods such as mathematics and statistics to describe, evaluate, and monitor research in a specific field in order to reveal the research structure and trends in a certain discipline (22, 23). In recent years, using CiteSpace for literature and metrological analysis has become a research hotspot for scholars at home and abroad. CiteSpace is a literature metrological modeling software that can be used for basic literature analysis, such as citation analysis, international and institutional cooperation analysis, author cooperation analysis, dual-map overlay of journals, keywords analysis, and clustering and mutation analysis. Through analysis, it provides insights into the structure, social network, and topic interests of the field (24, 25).

Hence, this study conducted a comprehensive review of publications in the field of post-stroke dysphagia research and used CiteSpace for bibliometric analysis. Based on the analysis results, we could help researchers quickly understand the main progress, research hotspots, and frontiers of post-stroke swallowing dysfunction, addressing the lack of quantitative analysis in this field.

Materials and methods

Data source and search strategy

The Web of Science database encompasses a diverse range of disciplines, with its fundamental principles encompassing data structuring, subject categorization, citation analysis, an international perspective, and scalability. It is the preeminent database utilized for bibliometric analysis (26). Earlier studies have convincingly established the efficacy of bibliometric analysis conducted on the WoSCC database (27, 28). The literature data relevant to the topic of this bibliometric study were obtained and retrieved from the core collection database in the Web of Science (WoS). Searches were conducted using the following MeSH terms: (((swallowing) OR (dysphagia)) OR (swallowing disorder)) AND (stroke). Our literature search was limited to the time period between 1 January 2013 and 31 December 2022, with a yearly time slice. This approach aims to prevent potential information obsolescence and degradation in data quality associated with long-term datasets, ensuring the timeliness and reliability of the retrieved information. Additionally, limiting the search duration to a decade enhances the efficiency and feasibility of data analysis and visualization, ultimately reducing the complexity and cost associated with data processing.

Inclusion and exclusion criteria

After a thorough examination of the article titles and abstracts, only those related to post-stroke dysphagia were selected for this bibliometric analysis. Other document types, such as meeting abstracts, letters, editorial materials, and book chapters, were excluded. Publications written in English were the only ones considered for this analysis. Figure 1 shows the flow chart of the selection process. In the end, a total of 1,447 records were deemed suitable and included in the final bibliometric analysis.

Software parameter settings

CiteSpace V (version 6.2.R4; Drexel University, United States) and Microsoft Excel 2019 were used for bibliographic analysis, with the "Time Sliding" value set to 1 year and the type of node selected based on the analysis's purpose. In this study, we employed "country, institution, author, reference, keyword" as node types for visual analysis, and only one node type can be selected at a time.

Interpretation of main parameters

Node circle and the link between nodes

The size of a node circle in the countries or institutional co-authorship network represents the number of papers published as well as the frequency of authors in the co-occurrence network. Meanwhile, a link between nodes shows the presence of a co-authorship or co-occurrence relationship.

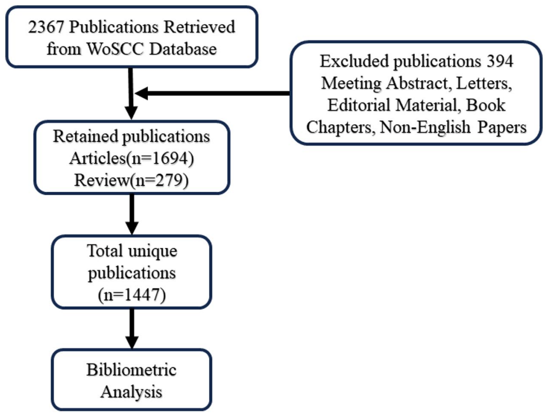

FIGURE 1
Flowchart of literature inclusion.

Betweenness centrality

The index of betweenness centrality is an indicator of the importance of nodes in a network.

Dual-map overlaps

The dual-map overlaps of journals are a novel method of visualizing the distribution of articles and citation trajectories across different fields. It provides insight into interdisciplinary relationships in the academic world. The citation line, represented by a curve, presents the context of the citation in a comprehensive manner. The use of the Z-score and F-score allowed for re-adjustment and standardization of the citation data, enabling the identification of major citation paths on the dual-map.

Burst detection

The purpose of burst detection is to identify significant increases in citation numbers within a particular timeframe.

H-index

H-index is a metric used to evaluate academic success. It is calculated by counting the number of publications that have been cited more than H times.

Results

Publication analysis

In this study, a total of 1,447 articles were included in Supplementary Figure S1 displays the annual publication and citation distribution of literature related to post-stroke dysphagia. We identified the overall and prominent sub-topic (clinical neurology) trend lines, represented by orange and blue colors, respectively. The orange line represents the general trend observed in a broader field, whereas the blue line highlights the specific trend in clinical neurology. As shown in Supplementary Figure S1A, it is evident that the number of publications in this field has been increasing, climbing from 72 (4.98%) in 2013 to 262 (18.11%) in 2022. Linear regression analysis indicates that the time trend of the number of publications in the past decade was significantly correlated ($R^2 = 0.9466$). However, it should be noted that the growth rate of clinical neurology was relatively slow. Supplementary Figure S1B demonstrates the distribution of citation frequency, indicating that the number of citations has been increasing year by year, and the growth of clinical neurology aligned with the overall growth pattern. By utilizing an exponential growth model to evaluate the correlation between the number of citations and the publication year, it was found that the model aligns with the observed trend.

Supplementary Figure S2 shows that 2022 recorded the most published papers ($n = 262$) and open access papers ($n = 189$). The highest number of citations per paper was 37.16 in 2016. The largest number of citations ($n = 3,456$) and the H index ($n = 31$) also occurred in 2016.

Authoritative journals analysis

A total of 1,447 papers on post-stroke dysphagia were published in 476 academic journals in this study. Table 1 summarizes the list of the top 15 academic journals, ranked by the number of publications. In the top 15 journals, dysphagia contributed the highest number of published articles ($n = 95$), and the greatest H-index value ($n = 23$), followed by the Journal of Stroke and Cerebrovascular Diseases ($n = 75$). Medicine and Stroke were tied for third place with 35 papers each. The International Journal of Stroke presented with the highest impact factor of 6.7 in 2022 and the largest quantity of citations per paper. The academic journal Frontiers in Neurology had the most open access ($n = 34$).

Dual-map overlaps of journals are shown in Figure 2. The map was assigned to two parts, with the citing journals listed on the left and the cited journals on the right. The Z-score function was used to highlight a more fluid trajectory, with higher scores represented by thicker lines. Five major citation trajectories were determined (pink and green), with journals in Medicine, Neurology, Sports, and Ophthalmology (pink trajectory) being more frequently cited by Psychology, Education, Social ($Z = 4.53$, $f = 4,638$), Health, Nursing, Medicine ($Z = 4.46$, $f = 4,572$), and Molecular, Biology, and Genetics ($Z = 3.98$, $f = 4,125$) fields. Additionally, journals in Medicine, Medical, and Clinical (green track) were influenced by journals in Health, Medicine, Medical ($Z = 3.83$, $f = 3,990$), and Psychology, Education, and Social ($Z = 1.98$, $f = 2,268$).

Subject category analysis

The 1,447 articles included in our study were classified into a total of 81 WoS subject categories. Supplementary Figure S3 shows the top 15 published disciplines ranked by the number of publications. Clinical neurology had the largest number of papers ($n = 338$), citations ($n = 6,814$), open-access papers ($n = 195$), and H-index value ($n = 43$). Peripheral vascular disease had the largest average number of citations per article ($n = 24.26$), followed by clinical neurology ($n = 20.16$) and nutrition dietetics ($n = 18.62$).

TABLE 1 Top 15 paper journals based on the number of publications.

Journal	Publications	Citations	Citation per paper	Open access	WoS categories	IF(2022)	Quartile	H-index
Dysphagia	99	1950	19.7	29	Otorhinolaryngology	2.6	Q2	23
Journal of Stroke and Cerebrovascular Diseases	75	954	12.72	16	Neurosciences; Peripheral Vascular Disease	2.5	Q3; Q3	17
Medicine	35	209	5.97	35	Medicine; General and Internal	1.6	Q3; Q3	8
Stroke	35	1,307	37.34	33	Clinical Neurology; Peripheral Vascular Disease	8.3	Q1; Q1	19
Frontiers in Neurology	34	245	7.21	34	Clinical Neurology; Neurosciences	3.4	Q2; Q2	9
Archives of Physical Medicine and Rehabilitation	21	329	15.67	4	Rehabilitation; Sport Sciences	4.3	Q1; Q1	11
Journal of Oral Rehabilitation	21	553	26.33	2	Dentistry, Oral Surgery & Medicine	2.9	Q2	13
Neurogastroenterology and Motility	20	461	23.05	6	Gastroenterology & Hepatology; Clinical Neurology; Neurosciences	3.5	Q2; Q3; Q2	12
Neurorehabilitation	18	188	10.44	2	Clinical Neurology; Rehabilitation	2	Q4; Q2	8
Annals of Rehabilitation Medicine	16	100	6.25	16	Rehabilitation	1.3	Q3	7
PLOS ONE	16	438	27.38	16	Multidisciplinary Sciences	3.7	Q2	10
BMC Neurology	15	154	10.27	15	Clinical Neurology	2.6	Q2	7
Cerebrovascular Diseases	15	375	25	7	Clinical Neurology; Peripheral Vascular Disease	2.9	Q3; Q3	10
European Journal of Neurology	15	337	22.47	3	Clinical Neurology; Neurosciences	5.1	Q1; Q2	10
International Journal of Stroke	15	842	56.13	5	Clinical Neurology; Peripheral Vascular Disease	6.7	Q1; Q1	9

WoS, Web of Science; IF, Impact Factor; JCR, Journal Citation Reports.

Authoritative countries, institutions, and authors analysis

A total of 84 countries contributed to the publication of research on post-stroke dysphagia. Supplementary Figure S4A shows the top 15 countries based on the number of publications, with China having the highest number of publications ($n = 182$) and open access papers ($n = 235$). The USA had the highest H-index ($n = 37$) and the most citations ($n = 4,871$). Switzerland had the highest number of citations per paper ($n = 48.12$), followed by Spain ($n = 29.28$) and England ($n = 24.75$). Supplementary Figure S4B shows the top 11 countries with the strongest citation bursts. Sweden had the highest burst strength, with a score of 4.61 from 2015 to 2018, indicating a significant focus on post-stroke dysphagia research in Sweden during this period.

Supplementary Figure S5A displays information on the top 15 institutions based on the number of research papers published. The University of London ranked first with 49 publications, followed by the University of Manchester with 44 and Harvard University with 37 publications. The University of Munster had the highest citation rate of 39.64 per paper. When analyzing the burstness of institutions in Supplementary Figure S5B, it was observed that the Veterans Health Administration (VHA) and the US Department of Veterans Affairs were tied for first place, both scoring 5 from 2013 to 2014. Dongseo University ranked third with a burst strength score of 4.07 from 2019 to 2020.

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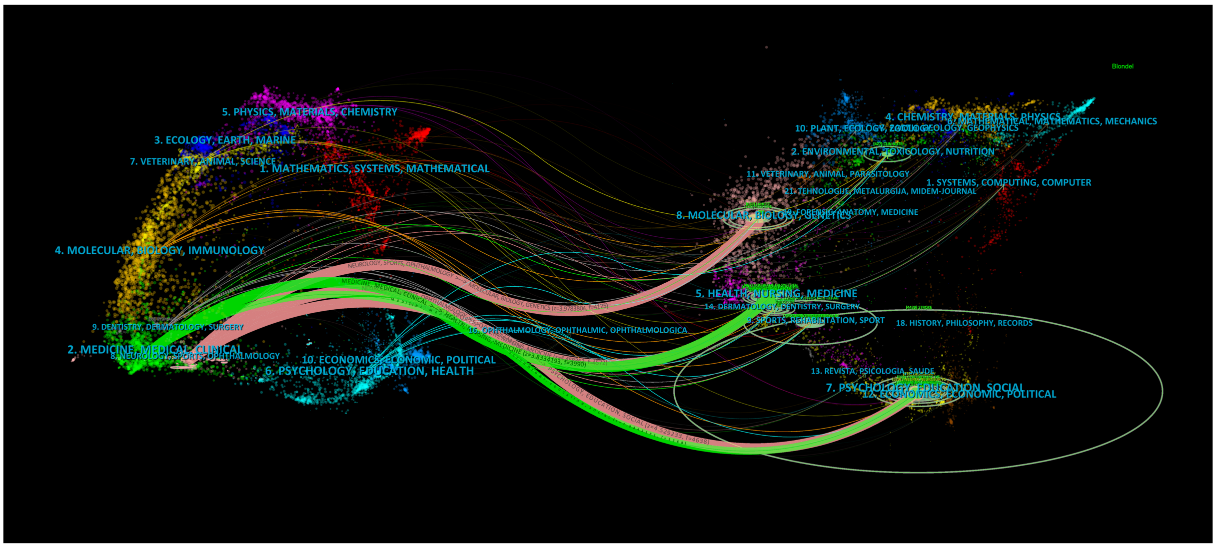

FIGURE 2
Dual-map overlaps of journals in post-stroke dysphagia. The map is assigned to two parts, with the citing journals listed on the left and the cited journals on the right.

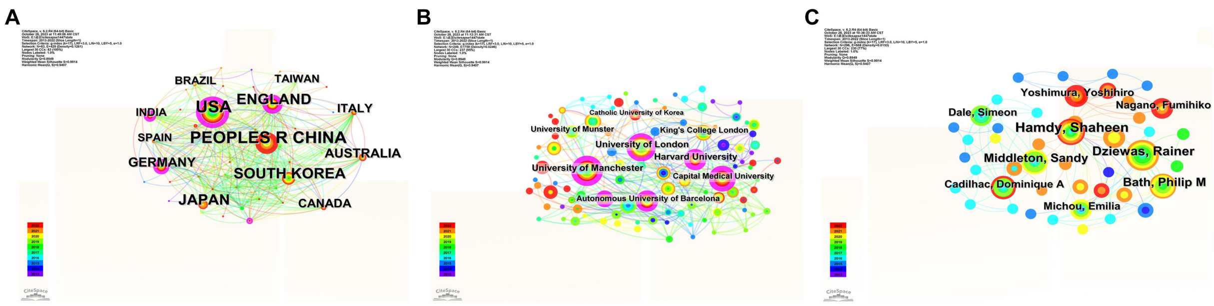

FIGURE 3
Close cooperation networks among countries **(A)**, institutions **(B)**, and authors **(C)** have been formed globally. Links indicate the presence of co-authorship or co-occurrence relationships.

Supplementary Figure S6A summarizes the top 15 most authoritative authors. Hamdy S had the largest number of papers (*n* = 25), with 22 of them being open access. Dziewas R had the highest number of citations (*n* = 915) and also shared the highest H-index (*n* = 15) with Clave P and Hamdy S. Additionally, Dziewas R had the greatest number of citations per paper, with a value of 45.75. Supplementary Figure S6B displays the top 15 authors with the strongest citation bursts. Park Ji-Su had the highest burst strength, scoring 4.12 from 2019 to 2020. Clave P had a score of 4.12 in burst strength, followed by Yoshimura Y with a score of 3.25 from 2020 to 2022. These scores indicated that both authors had a keen interest in the study of post-stroke dysphagia during this period.

Coauthorship analysis of countries, institutions, and authors analysis

Figure 3 displays the collaboration maps of countries, institutions, and authors. According to the total link strength, the top three countries were China (*n* = 273), USA (*n* = 251), and England (*n* = 145). The top three institutions were the University of London (*n* = 47), the University of Manchester (*n* = 44), and Harvard University (*n* = 37). As for authors, the top three authors were Hamdy S (*n* = 27), Dziewas R (*n* = 21), and Middleton S (*n* = 16) based on their contributions.

In terms of betweenness centrality, the United States had the highest centrality (*n* = 0.45), and the University of London and the University of Manchester were the top institutions with the highest centrality (*n* = 0.21). The top five authors were Hamdy S (*n* = 0.01), Dziewas R (*n* = 0.01), Middleton S (*n* = 0.01), Bath P (*n* = 0.01), and Michou E (*n* = 0.01).

Reference analysis

Figure 4 presents a timeline view of the references. The reference cocitation analysis grouped the research categories into 26 clusters (#0–26). The largest cluster (#0) consisted of 64 members and was classified as a controlled trial. The most relevant citation to this cluster

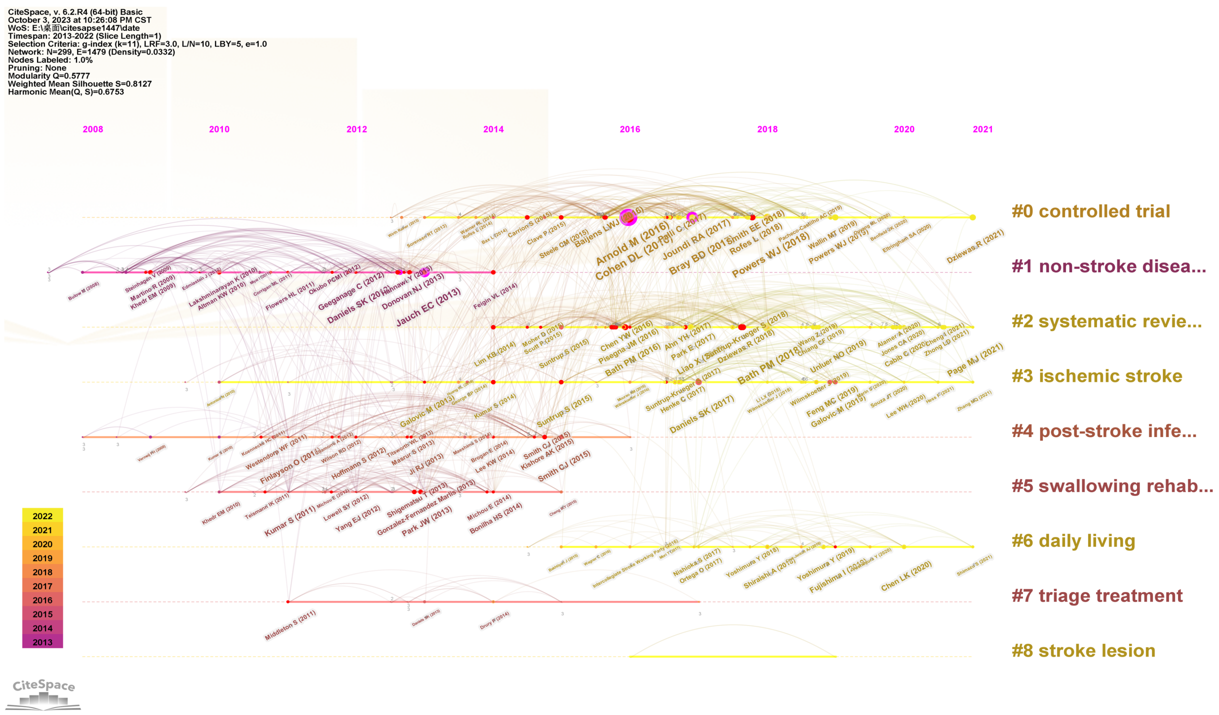

FIGURE 4
Timeline view of reference co-citation analysis.

was "European Stroke Organization and European Society for Swallowing Disorders Guideline for the Diagnosis and Treatment of Post-Stroke Dysphagia" (16). The second-largest cluster (#1), labeled as non-stroke diseases, contained 49 members. The most pertinent citation to this cluster was "Approaches to the Rehabilitation of Dysphagia in Acute Post-Stroke Patients" (29). The third-largest cluster was labeled as ischemic stroke, and the most relevant citation was "Predictors of Complete Oral Feeding Resumption after Feeding Tube Placement in Patients with Stroke and Dysphagia: A Systematic Review" (30).

Keyword analysis

Figure 5 displays the top 25 keywords with the strongest citation bursts. The keyword with the highest burst value was "implementation" ($n = 6.55$), followed by "predictors" ($n = 5.63$) and "systematic review" ($n = 5.08$). The keyword "classification" had the longest burst period, lasting from 2014 to 2018. As of the end of 2022, the most frequently cited keywords included "systematic review," "validity," "activities of daily living," and "swallowing disorders."

Discussion

Global research trends of post-stroke dysphagia

This study conducted a systematic and comprehensive bibliometric analysis of research on post-stroke dysphagia in the past 10 years using CiteSpace software. The results showed that the global trend of published papers on post-stroke dysphagia increased during the 10-year period from 2013 to 2022. However, it should be noted that the growth rate of clinical neurology was slower than the overall trend observed in this field. In addition, the global trend in citations increased from 44 to 5,405, with the most significant increase from 2019 to 2022. The increase in clinical neurology was consistent with the overall growth, indicating that dysphagia has received more attention in the field of stroke in recent years, with more and more scholars joining the research, further promoting the development of this field.

From the journal analysis, dysphagia was the Q2 journal with the highest publications and the largest citations. This indicates that publishing high-quality papers on post-stroke dysphagia is a challenge. In addition, among the top 15 journals, Stroke (IF, 2022 = 8.3), the International Journal of Stroke (IF, 2022 = 6.7), and the European Journal of Neurology (IF, 2022 = 5.1) were the Q1 journals with an IF score > 5. This indicates that the articles published in these three journals have a higher academic reference value and are more authoritative. Through analyzing the overlap of dual-maps in published journals to discern the citation trajectories across various fields, it becomes apparent that post-stroke dysphagia is influenced by a range of disciplines extending from medicine, neurology, and clinical health to psychology, education, and sociology. The disciplines mentioned above contribute to addressing this intricate problem from diverse angles, offering distinctive perspectives and approaches. Medicine and neurology concentrate on physiological mechanisms and therapeutic methods, clinical hygiene examines best practices, psychology emphasizes psychological wellbeing, education strives to enhance the disorder through educational means and training; and

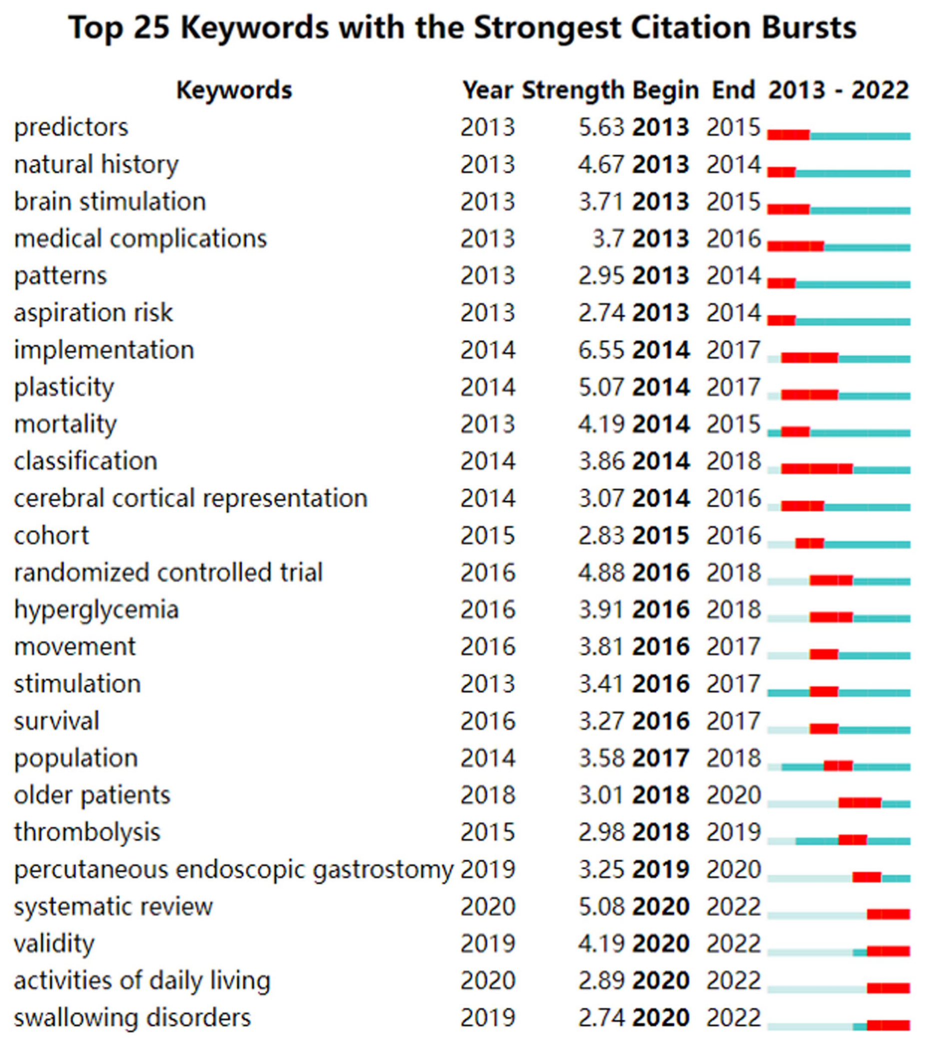

FIGURE 5
Top 25 keywords with the strongest citation bursts conducted by CiteSpace. The blue bars mean the reference has been published; the red bars mean citation burstness.

sociology emphasizes social engagement and support systems (16). Interdisciplinary collaboration and communication are essential for developing comprehensive treatment plans and enhancing patient outcomes in terms of quality of life and prognosis. Future research should take into account the dynamic nature of these field journals.

Among the top 15 authoritative countries ranked by publication quantity, two-thirds were developed countries, while only one-third were developing countries. In addition, although China had the highest number of papers, the United States had the highest number of citations and H-index, and Spain ranked first in citations per paper. This indicates that there are still significant disparities between developed and developing countries in the research on post-stroke dysphagia. This phenomenon may be due to the following reasons: First, some developing countries still focus their research on reducing the mortality rate of stroke, which is the second leading cause of death (31). Second, there are differences in the incidence rate of post-stroke dysphagia between developed countries and developing countries. Meta-analysis shows that the incidence rate in developed countries is approximately 44–61%, while that in developing countries is approximately 37–39% (11). The high incidence rate may prompt developed countries to pay more attention to post-stroke dysphagia. Finally, developed countries have more economic strength, medical investment, talent cultivation, and scientific research environment resources, which affect the output and quality of scientific research results in developed and developing countries. Among the authoritative institutions and authors, the University of London was the top-ranked institution, and Hamdy S was the most influential author. From the perspective of the collaboration network, the United States had the highest centrality. The University of London and the University of Manchester were the top institutions with the highest centrality. These results suggest that countries and institutions with the most publications do not necessarily have the highest degree of betweenness centrality. Future research involving post-stroke dysphagia should strengthen collaboration and cooperation between different countries and institutions to improve research quality.

Research hotspots and prospects of post-stroke dysphagia

The evolution of a knowledge field can be reflected through keywords. Therefore, keyword analysis can reveal research hotspots and development trends. According to keyword-based counting analysis, dysphagia ($n = 221$) ranked first, followed by aspiration ($n = 215$). Dysphagia is one of the most common complications after stroke (32, 33). Despite multiple advances in the treatment of hyperacute phase and secondary prevention of stroke, the treatment of post-stroke dysphagia remains a neglected research area (33–35). Swallowing and breathing share the pharynx and are both regulated by the medulla oblongata, making them two synchronized complex biomechanical processes (36, 37). During normal swallowing, the airway is closed, and breathing is paused to prevent food from entering the airway (38). Patients with dysphagia may have aspiration due to abnormal airway protection mechanisms, causing food to fall into the respiratory tract (39). Previous studies showed that in stroke patients, the incidence rate of dysphagia was consistent with pneumonia (11, 32). However, conventional diagnostic methods for dysphagia have limited accuracy in predicting aspiration and respiratory disease (40). In addition, there was insufficient randomized controlled trial data to determine the impact of screening programs for dysphagia on reducing post-stroke pneumonia (41). Therefore, more research is needed to compare the effectiveness of different screening methods for dysphagia in the future, and incorporating measurable objective assessments into clinical diagnosis is necessary. This may be the key to developing new treatment strategies. The keyword "classification" had the longest burst period. Warnecke et al. found that based on the flexible endoscopic evaluation of swallowing (FEES), the neurogenic dysphagia phenotype can be divided into seven categories (42). Stroke commonly manifests as "premature bolus spillage," "delayed swallowing reflex," "residual material in the piriform sinus," and "pharyngolaryngeal movement disorder" (12). Parkinson's disease often appears as "residual material in the valleculae" and "pharyngolaryngeal movement disorder" (43). Myasthenia gravis is commonly characterized by "fatigue-prone muscle weakness" (44). "Complex disorder" with a heterogeneous dysphagia pattern is more prevalent in amyotrophic lateral sclerosis (45). Therefore, the dysphagia phenotype is beneficial for the differential diagnosis of post-stroke dysphagia.

The subject category analysis showed that clinical neurology, neurosciences, and rehabilitation were the main categories of research on post-stroke dysphagia, indicating that research on post-stroke dysphagia from the perspective of clinical neurology focuses on rehabilitation. In the reference analysis, the most relevant citer to the cluster "swallowing rehabilitation "was "Transcranial non-invasive brain stimulation in swallowing rehabilitation following stroke--a review of the literature" (46). This study found that, based on available evidence, non-invasive brain stimulation may provide a useful adjunctive therapy for post-stroke dysphagia rehabilitation. In addition, according to the timeline perspective of literature analysis, "daily living" has also been a hot topic in recent years. Dysphagia may prevent individuals from living independently and returning to work due to conditions such as being unable to eat orally, using nasal feeding tubes, developing aspiration pneumonia, experiencing malnutrition, and suffering from mental disorders, which significantly impact the quality of life (47). Therefore, more high-quality research on non-invasive interventions for post-stroke swallowing dysfunction is needed in the future to improve the quality of life of patients with post-stroke dysphagia.

Although this study reveals the trends and hotspots of post-stroke dysphagia, there are still some topics and areas that have not been fully studied. For example, the efficacy of swallowing exercises and rehabilitation programs, the specific needs of elderly patients, and the application of technology in diagnosis and management (39, 48–50). In-depth research on these topics will help medical practitioners better understand post-stroke dysphagia and provide more effective treatment and management strategies.

Strengths and limitations

As far as we know, this study represents the first bibliometric and visual analysis of post-stroke dysphagia, drawing from literature published between 2013 and 2022. In addition, this study reviews the progress and trends of research on post-stroke dysphagia worldwide. However, it should be noted that there are several limitations. First, considering the limitations of CiteSpace software and in order to ensure the quality of the retrieved publications and the integrity of the information, we only searched the core collection of Web of Science, which may have omitted important literature from other databases. Second, in the search strategy, we did not incorporate all keywords associated with dysphagia. To achieve a more comprehensive understanding of this field, future research can refine the search strategy based on this foundation, leading to more robust evidence support. Finally, our study only included publications from 2013 to 2022 and did not incorporate the most recently published high-quality papers. This may lead to an incomplete representation of the latest research dynamics and advancements, particularly in rapidly evolving fields such as post-stroke dysphagia.

Conclusion

In conclusion, this study may help researchers reveal the publication patterns and emerging trends of post-stroke dysphagia from 2013 to 2022. The most influential authors, institutions, journals, and countries were Hamdy S, University of London, Dysphagia, and the People's Republic of China. The visual map displays the hot research directions of post-stroke dysphagia studies in recent years, including dysphagia and aspiration, dysphagia classification, dysphagia rehabilitation, and daily living. In stroke patients with dysphagia, accurate identification of the type of swallowing disorder and in-depth exploration of novel rehabilitation techniques, such as non-invasive brain stimulation, contribute to the development of more scientific and effective treatment protocols. Furthermore, precise assessment of the risk of aspiration and the implementation of appropriate nursing interventions contribute to reducing the risk of pneumonia and, ultimately, significantly enhancing the patients' quality of life. Therefore, it is crucial to conduct large-sample randomized controlled trials on screening programs and non-invasive intervention methods for post-stroke dysphagia in the future.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding authors.

Author contributions

BG: Data curation, Writing – original draft. ML: Writing – review & editing. ZW: Conceptualization, Writing – review & editing. ZY: Conceptualization, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

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Quantitative evaluation of swallowing function in Parkinson's disease using tongue pressure measurement: a mini-review

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Dysphagia is a common symptom of Parkinson's disease (PD) associated with aspiration pneumonia, choking, malnutrition, and a decreased quality of life, and is a leading cause of death among patients with PD. Tongue dysfunction in patients with PD affects the oral phase of swallowing, including the formation and propulsion of a bolus into the pharynx. Assessing tongue pressure, generated between the tongue and palate, is a method that quantitatively measures tongue function and is related to dysphagia in PD. Two assessment methods are used to measure tongue pressure: tongue strength and tongue pressure during swallowing. Previous studies measuring tongue pressure in PD have reported decreased tongue strength and pressure during swallowing, as well as a prolonged tongue pressure rise time, which are symptoms associated with PD severity and dysphagia. In this mini-review, we present a method for measuring tongue pressure and discuss its relationship with dysphagia in PD. We also describe limitations and future perspectives in tongue pressure measurement research.

KEYWORDS

Parkinson's disease, dysphagia, tongue pressure, tongue strength, swallowing

1 Introduction

Dysphagia occurs frequently in Parkinson's disease (PD), not only leading to a decline in the quality of life related to meals, depression, and malnutrition, but also serving as a cause of aspiration pneumonia, which is associated with life prognosis (1–6). In PD, bradykinesia, hypokinesia, and tremors can also influence swallowing organ motility and cause problems in the oral phase, in which a high prevalence of abnormal tongue movement is typically observed (7, 8). Tongue tremors, pumping-like movements, prolonged tongue elevation, and muscle weakness are also observed, and these motor abnormalities can cause difficulty initiating swallowing, prolonged oral transit time, difficulty propelling a bolus from the oral cavity to the pharynx, a decreased propulsive force, and oral residuals (9, 10). Given these considerations, it is important to establish a clinical marker of abnormal tongue movement in the evaluation of dysphagia in PD.

The tongue plays an important role in propelling a food bolus from the oral cavity to the pharynx during swallowing, and the decreased production of tongue pressure is a risk factor for impaired safety and efficiency during swallowing (11). Tongue pressure, a measure of the pressure produced between the tongue and palate, is an indicator of tongue motility. In patients with PD, abnormal tongue pressure patterns and movements during swallowing have been reported. Measuring tongue pressure is a less invasive assessment method and may have diagnostic value in the evaluation of dysphagia in patients with PD. Given this background, the present mini-review aimed to summarize the research literature on the measurement of tongue pressure in patients with PD. A literature search was performed on PubMed, Web of Science, and Google Scholar using the terms "Parkinson's disease" AND "tongue pressure" OR "lingual pressure" OR "tongue strength." We reviewed the title and abstract of each result and selected articles related to tongue pressure studies in patients with PD.

2 Types of methods for measuring tongue pressure

There are two types of tongue pressure: maximal tongue pressure, which occurs when the tongue is voluntarily pushed strongly upward, and tongue pressure during swallowing, which is produced between the tongue and palate during swallowing. Researchers have used various terms to refer to maximal tongue pressure, including tongue strength, lingual strength, maximal lingual pressure, maximal tongue pressure, and tongue pressure strength (12–15). Because both types of tongue pressure are performed using maximal isometric movement, they are used to indicate tongue strength. In this paper, to avoid confusion among readers, the term tongue strength is used throughout.

Tongue pressure during swallowing is a method of measuring the pressure, location, and timing of tongue contact with the palate by a sensor placed in the intraoral cavity during the swallowing of saliva, food, or drinks.

3 Measurement of tongue strength

3.1 Measurement instruments and methods

Tongue strength is measured using a tongue pressure measuring device that consists of an instrument unit, a connecting tube, and a tongue pressure probe. A probe with a balloon-shaped tip is placed on the tongue in the oral cavity and pressed with maximal force against the palate to measure the pressure produced. The subject is instructed to "press as hard as you can with your tongue against the plastic bulb." Measurements are taken three times and the average or maximal value is recorded as the representative value for each subject. A typical tongue pressure measuring device is the IOPI Pro (Model 3.1; IOPI Medical LLC, Woodinville, WA, United States), which is the most widely used device and has been the subject of many research reports (16–19) (Figure 1A). The IOPI is a handheld portable device that uses an air-filled plastic bulb (3.5 cm long; 4.5 cm diameter, 2.8 mL internal volume) (20). Another common device is the JMS Tongue Pressure Measurement Device (TPM-02; JMS Co., Hiroshima, Japan), which is used mainly for clinical and research purposes in Japan (21–25) (Figure 1B). The tip of the IOPI probe has a slightly flattened shape, whereas that of the JMS device has a nearly spherical shape. The JMS device has a rigid ring at the base of the balloon that is secured by the incisors to control mandibular movement and balloon positioning. Because the IOPI probe has no fixed points on the incisors, the probe position can be moved from the anterior to posterior portion of the tongue. Both the IOPI and JMS instruments are expressed in kilopascals (kPa), but because the instruments are different, the readings are not directly comparable. A conversion formula for the relationship between the tongue strength value of both devices has been reported by Yoshikawa et al. (26). Real-time and tongue strength value displays are common features of both devices.

These instruments can also assess endurance by measuring not only tongue strength, but also the time it takes to keep the tongue pressed with a constant force. Tongue endurance is measured by the time that 50% of maximal tongue strength can be sustained with feedback of the pressure ramp displayed on the tongue pressure measuring device and the waveform displayed on the monitor using specialized software (27, 28).

These tongue pressure measuring devices can also be used for tongue strength training, and have been applied to rehabilitation therapy using tongue strength values as an indicator.

3.2 Tongue strength in PD

A number of studies have shown that patients with PD have decreased tongue strength and endurance compared with controls in similar age groups (29–31). Solomon et al. (30) investigated muscle strength and endurance in the tongue and hands of patients with PD and compared them with neurologically normal controls, and reported that the patients with PD had 8.3 kPa lower tongue strength and 8.2 s shorter endurance. Tongue strength can be measured in the anterior and posterior portions of the tongue, but studies of PD have reported declines in only the anterior portion or in both the anterior and posterior portions (32, 33).

Regarding the relationship with the progression of PD, tongue strength has been shown to decrease more in patients with advanced than in patients with mild/moderate disease progression. Plaza et al. (34) reported a negative correlation between Hoehn and Yahr stage and tongue strength, which decreases with the progression of PD. They reported finding no differences in tongue endurance or gender based on the degree of PD progression. According to a meta-analysis by Pitts et al. (31), decreased muscle strength and endurance in the anterior part of the tongue is expected in approximately one-third and one-fourth of patients with PD, respectively. They point out that the decrease in the anterior part of the tongue appears from stage II of the Hoehn and Yahr classification and may become more persistent as the disease progresses.

An analysis of the relationship between tongue pressure and swallowing function using videofluoroscopic swallowing studies reported that higher tongue strength leads to less airway penetration of thin liquids, and that low endurance in the anterior portion of the tongue delays the laryngeal vestibule closure time (33, 35).

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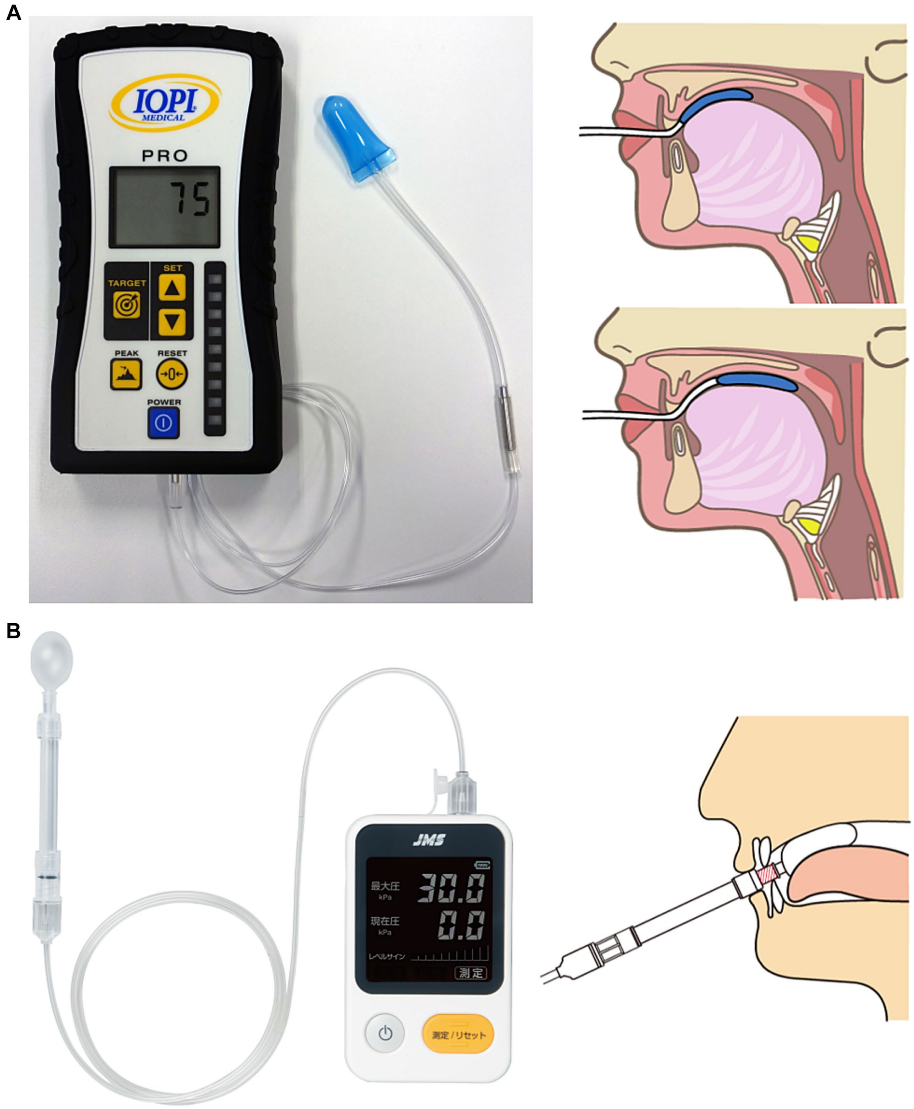

FIGURE 1
(A) IOPI Pro (Model 3.1) and measurement methods. **(B)** JMS Tongue Pressure Measurement Device (TPM-02) and measurement methods.

Weak tongue strength is also associated with subjective symptoms of dysphagia. Tongue strength is decreased in patients with PD with subjective symptoms of dysphagia compared with those without PD (36). Pitts et al. (32) reported that patients with PD with reduced muscle strength in the anterior part of the tongue had lower total scores on the Swallowing Quality of Life (SWAL-QOL) questionnaire and reported subjective symptoms such as prolonged eating time and decreased motivation to eat.

De Letter et al. (37) examined the effect of levodopa on tongue pressure. Ten patients with idiopathic PD were studied for isometric motor tasks of the tongue in the on and off phases. The maximal force of tongue movement and contraction time were not significantly different between the two conditions, but the integral (area under the curve) was significantly greater in the on phase. In addition, the force decay slope was significantly lower in the on phase. These findings are consistent with the pathophysiological effects of isometric muscle contraction patterns in PD.

4 Measurement of tongue pressure during swallowing

4.1 Measurement instruments and methods

Tongue pressure during swallowing is a method of measuring the contact pressure between the tongue and palate during swallowing. A simple method is to place the balloon of a tongue-pressure measuring device such as the IOPI on the tongue and measure the pressure of the tongue pushing the balloon upward during swallowing (33, 35). A detailed evaluation of tongue pressure during swallowing involves attaching a customized sensor to the palate and measuring the contact between the tongue and the palate. Multiple pressure sensors are installed in the palate, including in the bulb and mouthpiece (38–40). The tongue pressure sensor (Nitta Co., Osaka, Japan) has five pressure-sensitive sensors arranged in an ultra-thin sheet (0.1 mm thick), allowing it to measure tongue pressure during swallowing under natural conditions with minimal discomfort (39, 40). The measurement of tongue pressure during swallowing provides detailed objective information on the site of tongue contact with the palate, the order of tongue pressure onset, and the maximal tongue pressure, duration, and integral value for each site.

4.2 Tongue pressure during swallowing in PD

Studies using IOPI bulbs have shown that tongue pressure during salivary swallowing is decreased in patients with PD compared with healthy older adults (11, 35). Da Costa et al. (33) compared tongue pressure during salivary swallowing in 23 patients with idiopathic PD (mean age, 64.9 years) and 24 healthy controls (mean age, 64.1 years) using the IOPI placed anteriorly and posteriorly. They reported that compared with the control group, patients with PD had significantly lower tongue pressure during swallowing in the posterior region.

Hadley et al. (38) measured real-time tongue-palatal pressure in patients with PD using an oral mouthpiece with seven implanted pressure sensors. Using this device, they could distinguish between swallowing tasks of various samples, including saliva and water, and non-swallowing tasks, such as singing, chewing, speech, and isometric tongue push-up movements.

Minagi et al. (41) measured tongue pressure during swallowing in patients with PD using an ultra-thin sensor sheet with five pressure sensors affixed to the hard palate. The maximal tongue pressure at the measurement point was significantly lower in patients with PD than in healthy controls. Maximal tongue pressure was lower in patients with PD with dysphagia than in patients with PD without dysphagia. Loss of tongue pressure production in the anterior region of the hard palate was strongly associated with dysphagia in the oral and pharyngeal phases. They reported that abnormal tongue pressure production patterns, including partial or complete loss of tongue pressure, were observed at a higher rate in patients with PD with dysphagia than in patients with PD without dysphagia. They concluded that measuring tongue pressure during swallowing can detect not only the changes associated with overt dysphagia, but also the decreased tongue movement present in subclinical dysphagia.

Fukuoka et al. (42) used the same sensor sheet as Minagi et al. (41) to examine the characteristics of tongue movement in patients with PD (Figure 2). They compared tongue pressure during swallowing (maximal magnitude, duration, time-to-peak pressure, and pressure gradient) in dysphagia and non-dysphagia groups based on the findings of a videofluoroscopic swallowing study. No difference in maximal pressure was found between the two groups, but the duration and time-to-peak pressure were prolonged and the pressure gradient was decreased. These parameters may indicate temporal abnormalities in tongue movement in patients with PD. They concluded that measuring tongue pressure during swallowing using a tongue pressure sensor sheet can detect abnormal tongue movements in patients with PD, and is therefore useful in the diagnosis and treatment of dysphagia.

Other devices that may be able to measure tongue pressure during swallowing include the KayPentax 3-bulb array or the Madison Oral Strengthening Therapeutic device, but no data were found from studies with patients with PD (12, 43).

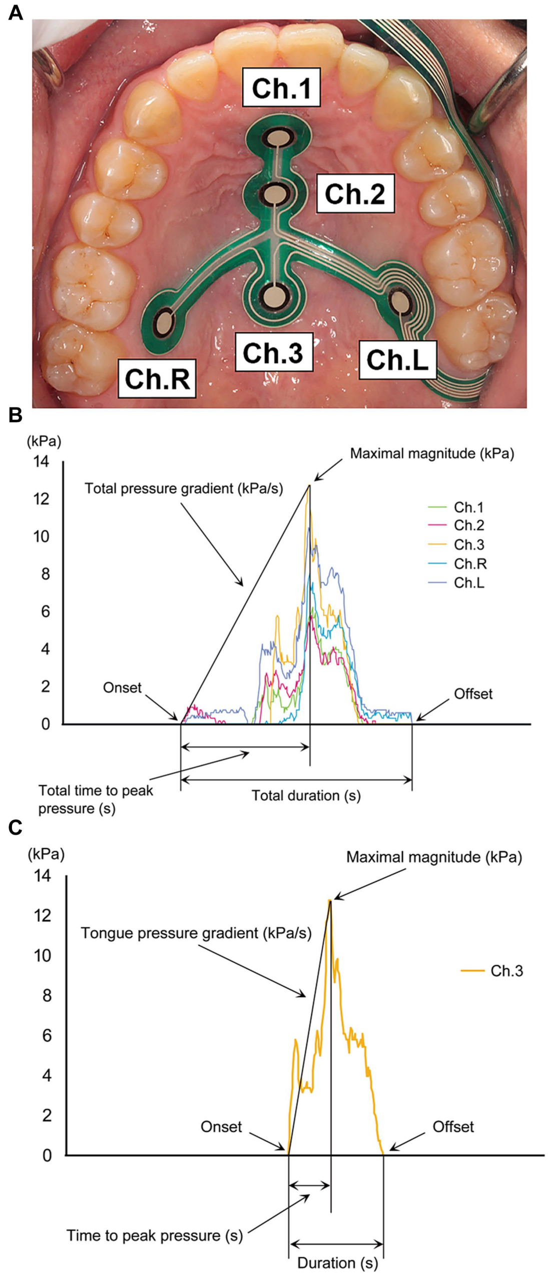

FIGURE 2
(A) Tongue pressure sensor sheet on the hard palate with five pressure-sensing parts. Ch.1 = anterior-median part; Ch.2 = mid-median part; Ch.3 = posterior-median part; Ch.R = right circumferential part; Ch.L = left circumferential part. **(B)** Tongue pressure waveforms of all channels recorded during swallowing. **(C)** Items for measuring tongue pressure during swallowing in a single channel.

5 Discussion

Most patients with PD have disorders related to tongue pressure, and tongue strength and endurance, as well as tongue pressure during swallowing, are known to be related to swallowing efficiency and safety (11, 29–32, 34, 41, 42). Tongue strength and endurance are reduced by physiologic factors other than aging and worsen as PD progresses (31, 34). Decreased tongue strength and endurance in PD may be related to the cardinal features of basal ganglia dysfunction, such as bradykinesia and hypokinesia (32). Muscle strength, especially in the anterior part of the tongue, is retained in the early stages of PD (31), but declines in the more severe stages and may be a leading indicator with respect to sensitivity to disease progression.

Tongue pressure is associated with subjective symptoms of swallowing and eating-related quality of life. Patients with PD with impaired tongue pressure have lower SWAL-QOL scores because of the effects on items such as eating duration, food selection, symptom frequency, and eating desire (32, 36). Because these subjective symptoms can be attributed to dysphagia, tongue pressure should be evaluated for the purpose of detecting dysphagia in patients with PD.

Data on muscle strength in the posterior part of the tongue have not been fully accumulated, and there is room for further study of its diagnostic significance. In addition, there are few reports of studies on tongue endurance compared with tongue strength. Although there have been reports suggesting an association between decreased anterior tongue endurance and delayed laryngeal vestibular closure, the relationship between tongue endurance and swallowing function remains unclear (33). Investigating the effects of reduced muscle strength and endurance on swallowing efficiency and safety for the anterior and posterior portions of the tongue, respectively, could clarify whether tongue pressure is a clinical marker for the presence or absence of dysphagia and the pathophysiology of PD. Because tongue pressure measuring devices can provide visual feedback in the form of numerical values and waveforms, they are expected to be used in rehabilitation therapy, such as for strengthening the tongue muscles. Several studies in older adults and stroke patients with dysphagia have reported increased tongue strength, reduced pharyngeal residuals, decreased aspiration, and improved quality of life related to swallowing function following tongue strength training (14–16, 44–46). In the future, it will be necessary to study the effects of improved tongue strength and endurance through rehabilitation on improved swallowing function, subjective symptoms, and quality of life.

Measuring tongue pressure during swallowing is a method of assessing tongue–palate contact pressure during the swallowing of food and drinks, and evaluates tongue dynamics differently from measurements of tongue muscle strength and endurance. Tongue pressure during swallowing has been measured by various devices in patients with PD, all of which show a decrease compared with healthy older adults of the same age. It is important to note that the bulb-type measuring device only measures the tongue–palate contact pressure at one location during swallowing. In addition, because the patient swallows with the bulb in place, there is added resistance to tongue movement, which may induce unusual swallowing dynamics. The sensor sheet and experimental palatal plate are shaped to fit the palate, which allows simultaneous measurement of multiple tongue–palate contact sites.

The measurement of tongue pressure during swallowing using a sensor sheet has revealed abnormal tongue movement in patients with PD. Compared with healthy older adults and patients with PD without dysphagia, patients with PD with dysphagia have partial or complete deficits in tongue pressure during swallowing and prolonged tongue–palate contact time and time-to-peak pressure (41, 42). Patients with PD tend to have abnormal tongue pressure during swallowing, which is important for propelling a food bolus from the oral cavity to the pharynx, as the efficiency of swallowing is reduced. These features identified in the tongue pressure waveform are consistent with the dysphagia findings in videofluoroscopic swallowing studies, as well as in the poor and uncoordinated food feeding movements in the oral phase of swallowing in patients with PD (7, 9, 10, 47).

Measuring tongue pressure during swallowing may provide an assessment of how the normal pattern is disrupted compared with normal subjects based on changes in the order of tongue pressure onset, duration, and maximal magnitude. If the changes in swallowing-related organ movements can be quantitatively assessed with high sensitivity, it may be possible to identify and effectively respond to dysphagia in patients with PD from the early stage.

A remaining challenge is that most devices that measure tongue pressure during swallowing were developed for research purposes or are not commercially available. Direct comparisons or conversions of pressure measurements between these devices has not yet been established. In the future, it will be necessary to develop commercial instruments that can be shared by researchers and to integrate existing data for further analysis.

Tongue pressure measurement is an excellent assessment of tongue strength and tongue-palate contact during swallowing. However, it is challenging to measure involuntary movements, such as resting tremor and dyskinesia, which are characteristic of PD patients. A comprehensive evaluation of swallowing function requires a multifaceted examination that extends beyond tongue pressure alone. Integrating tongue pressure measurement into clinical observation and imaging evaluations may enable a more detailed analysis of swallowing function in PD patients.

Author contributions

TF: Conceptualization, Funding acquisition, Investigation, Methodology, Writing – original draft, Writing – review & editing. KH: Conceptualization, Investigation, Methodology, Writing – original draft. TO: Conceptualization, Supervision, Writing – review & editing.

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Conflict of interest

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Complications of oropharyngeal dysphagia in older individuals and patients with neurological disorders: insights from Mataró hospital, Catalonia, Spain

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Background: Oropharyngeal dysphagia (OD) significantly impacts older individuals and neurologically compromised patients, hindering safe ingestion of food and liquids. Despite its prevalence, OD remains underdiagnosed and undertreated, leading to severe complications such as malnutrition, dehydration, respiratory infections, and aspiration pneumonia (AP), and increases hospital readmissions.

Objectives: This study analyzes the intricate relationship between OD and various clinical complications in older individuals and patients with neurological disorders.

Methods: Utilizing retrospective analysis and narrative review, our work consolidates findings from prior studies on Hospital de Mataro's dysphagia patient cohort. Revisiting OD's intricate association with clinical complications, it presents data via odds ratios (OR), incidence ratios (IR), and hazard ratios (HR) from univariate and multivariate analyses.

Results: Five studies (2001–2014) involving 3,328 patients were scrutinized. OD exhibited independent and significant associations with various complications among older patients. Older individuals with OD faced heightened 1-month (ODDS 3.28) and 1-year (OR 3.42) mortality risks post-pneumonia diagnosis. OD correlated with a 2.72-fold risk of malnutrition, 2.39-fold risk of lower respiratory tract infections, 1.82-fold pneumonia readmissions (IR), and 5.07-fold AP readmissions (IR). Post-stroke OD is linked to neurological impairment (OR 3.38) and respiratory (OR 9.54) and urinary infections (OR 7.77), alongside extended hospital stays (beta coefficient 2.11).

Conclusion: Oropharyngeal dysphagia causes and significantly exacerbates diverse clinical complications in older and post-stroke patients, emphasizing the urgent need for proactive identification, comprehensive assessment, and tailored management. Acknowledging OD's broader implications in general medical practice is pivotal to improving patient outcomes and healthcare quality.

KEYWORDS

oropharyngeal dysphagia, swallowing disorders, aspiration pneumonia, clinical complications, post-stroke

Background and aims

Dysphagia encompasses a spectrum of conditions that includes both oropharyngeal and esophageal dysphagia. These two forms, while falling under the same umbrella term, have distinct characteristics and implications. However, the scope of this article is specifically confined to oropharyngeal dysphagia and any subsequent reference to "dysphagia" in this article should be understood as referring exclusively to oropharyngeal dysphagia, unless explicitly stated otherwise. Oropharyngeal dysphagia is a multifaceted medical condition with significant health implications for various patient groups. It emerges from complex interactions between neurology, gastroenterology, and related fields, extending beyond mere swallowing difficulty to present a broad challenge with wide-ranging clinical implications. Regrettably, dysphagia is often overlooked, misdiagnosed, or underestimated in general medical practice, leading to a series of clinical complications that demand immediate attention (1).

The reported prevalence of dysphagia in published literature varies greatly due to different diagnostic methods. Studies indicate that dysphagia affects a substantial portion of individuals aged 70 years or older, with estimates ranging from 27 to 91% (2, 3). For patients with neurological conditions, the prevalence can be as high as 50% (1, 3). This positions dysphagia as a key concern in geriatric care (4). Complications of dysphagia, such as aspiration pneumonia, are particularly worrisome. One study found that about 33% of older patients with dysphagia developed aspiration pneumonia during rehabilitation, while another reported a 48.2% prevalence in a cohort of patients with dysphagia (5, 6). Although estimates vary, it is generally recognized that individuals with dysphagia face a higher risk of aspiration pneumonia compared to those without swallowing difficulties. Cabre et al. (7) established a strong link between dysphagia and pneumonia in older patients, emphasizing the prognostic implications and increased risk of readmission due to dysphagia. Dysphagia is also closely associated with malnutrition, with studies indicating a significant prevalence ranging from 25 to 45% in various patient cohorts (8, 9). Those with dysphagia often struggle to consume food properly, leading to decreased nutrient intake and potential deficiencies in essential nutrients. This research has advanced our understanding of dysphagia as a precursor to malnutrition and respiratory tract infections, adding complexity to its management in independently-living older individuals.

Besides its impact on health and quality of life, untreated or poorly managed dysphagia and its complications present a significant economic burden (10–12). Research at Mataró Hospital found that post-stroke patients with dysphagia incurred notably higher costs at various stages than those without dysphagia during hospitalization (€5357.67 vs. €3976.30), 3 months post-hospitalization (€8242.0 vs. €5320.0), and 12 months' follow up (€11,617.58 vs. €7242.78) (13). Importantly, patients with dysphagia who were at risk of malnutrition or malnourished and suffered respiratory infections incurred even higher costs at 12 months' follow up (€19,817.58 vs. €7242.8) (13). These findings highlight the substantial financial impact of dysphagia, especially when coupled with complications, and emphasize the potential benefits of early intervention and effective management in reducing these costs while improving patient outcomes.

The wider significance of these findings is underscored in key reviews, leading to a broader understanding of dysphagia as a geriatric syndrome. Baijens et al.'s white paper, backed by the European Society for Swallowing Disorders and the European Union Geriatric Medicine Society, articulates this concept well (4). However, this research is just one aspect of a larger issue that requires collective recognition and action. Many caregivers, both informal and formal, may lack awareness and knowledge of dysphagia, resulting in less than optimal care practices (14–18). There is a pressing need for targeted education and training programs to enhance dysphagia recognition, safe feeding practices, and strategies for managing swallowing difficulties. While this remains a significant challenge, the use of artificial intelligence and innovative interventions like the Minimal-Massive Intervention offer a promising shift in dysphagia diagnosis and management (19, 20). Minimal-Massive Intervention employs advanced, less invasive, compensatory strategies, and oral health to achieve substantial therapeutic benefits with minimal physiological disruption. By providing caregivers with the necessary knowledge and skills, including awareness of innovative approaches like Minimal-Massive Intervention, we can optimize care for individuals with dysphagia and reduce associated risks and costs.

This narrative review highlights the crucial issue of limited awareness and understanding of dysphagia complications among caregivers, including general practitioners. Clinical complications resulting from delayed recognition and misdiagnosis can lead to malnutrition, aspiration pneumonia, and compromised quality of life. Through an in-depth review of comprehensive studies conducted at a single center, we aim to highlight the urgent need for proactive identification, tailored management, and increased awareness to enhance the diagnosis and management of dysphagia.

Methods

In this review, we collected and compiled data from five separate investigations conducted by the same research team at Mataro Hospital between 2001 and 2014 (7, 20–23). These studies were carefully selected due to their shared location, consistent methodologies, and expert management. This selection aimed to ensure a uniform and cohesive research portfolio, enhancing the reliability of our findings for meaningful and comprehensive insights.

Study selection and rationale

Each of the five studies was chosen based on their specific focus and shared attributes, ensuring a common purpose and execution. The key factors guiding their selection were:

1. Location and facility: all studies took place at Mataro Hospital conducted by the same research team, providing a standardized clinical and research environment for practices and methodologies.
2. Methodological consistency: a deliberate effort was made to select studies with consistent research methods, allowing for easier cross-referencing and comprehensive understanding.
3. Expert assessment: the studies were managed by well-recognized and experienced professionals with deep understanding of the subject matter, ensuring the reliability of the results.

Methodological uniformity

Five studies examined dysphagia prevalence, risk factors, interventions, and complications in clinical settings like post-stroke, pneumonia, and older hospitalized populations. They all used the volume-viscosity swallow test, a validated tool for dysphagia diagnosis, assessing swallowing safety and efficacy with varying substance volumes and viscosities, and detecting silent aspirations via pulse-oximetry. Comprehensive evaluations were conducted on factors such as comorbidities (Charlson Comorbidity Index), functional abilities (Barthel Index), and nutritional status (Mini-Nutritional Assessment short-form). This uniformity allowed for a seamless approach within each study, contributing to coherence in the findings and enabling the research team to derive comprehensive conclusions from each individual study.

Data extraction

Our data extraction process involved a detailed review of five studies conducted at Mataró Hospital, chosen for their shared location, consistent methods, and expert management. We have been using electronic medical records (TESIS/HCE; Nexus, Sabadell, Barcelona, Spain) since 2007, aiding in recording patient outcomes. This system facilitated the extraction process, which examined clinical databases and electronic notes for 3,328 patients in our dysphagia cohort. The researchers conducting the extraction were experienced and knowledgeable, ensuring the accuracy of the information. The data included variables like demographic details, clinical assessments, diagnostic tools like the volume-viscosity swallow test and videofluoroscopy, and outcome measures such as hospital readmissions and mortality rates. This detailed extraction process enhances our methodology's transparency and validates our study's findings.

Outcome measures

A core set of clinical outcomes, including hospital readmissions, lower respiratory tract infections, pneumonia, and 6-month mortality after discharge, were assessed consistently across all studies. This uniformity in outcome measures simplified the comparison of results and enabled the identification of overarching trends and patterns.

Outlined below are concise summaries of the five studies, each addressing specific aspects of the research question, collectively contributing to a comprehensive synthesis of findings:

- Study 1, a prospective cohort in an acute geriatric unit, focused on dysphagia in pneumonia patients aged over 70 (134 patients; 80 female; mean age 78) (21). Dysphagia prevalence, clinical status, and prognosis were evaluated using bedside assessments, water swallow tests, and scoring systems like the Barthel Index, Mini Nutritional Assessment, Charlson Comorbidity Index, and Fine's Pneumonia Severity Index. 55% showed dysphagia signs, correlating with older age, poorer functional status, higher comorbidity, and severe pneumonia. Dysphagia patients had higher 30-day (22.9 vs. 8.3%, $p = 0.033$) and 1-year mortality rates (55.4 vs. 26.7%, $p = 0.001$).
- Study 2 was a population-based cohort study, evaluating dysphagia as a risk factor for malnutrition and respiratory infections in 254 community-dwelling individuals aged 70 and over (118 female; mean age 78) with a 90% follow-up rate at 1-year (23). Dysphagia was assessed using the volume-viscosity swallow test, along with evaluations for malnutrition, hand grip, Barthel score, and lower respiratory tract infections. No significant difference in annual malnutrition risk was found between groups with or without dysphagia. However, prevalent malnutrition cases at follow-up were linked to baseline dysphagia and impaired swallow efficacy. Patients with impaired swallow safety had higher annual respiratory infection rates.
- Study 3 was an observational prospective cohort study, examining whether dysphagia is a determining factor for pneumonia-related readmissions in older patients discharged from an acute geriatric unit (7). Analyzing data from clinical databases and electronic notes of 2,359 patients (1,461 female, mean age 84.9 years) followed for an average of 24 months, the study found that dysphagia was diagnosed in 47.5% of cases. Individuals with dysphagia had a higher incidence rate of readmissions for pneumonia—6.7 readmissions per 100 person-years compared to 3.67 in those without dysphagia. Dysphagia correlated with a higher risk of hospitalization for pneumonia (hazard ratio 1.6), with significantly increased risks for both aspiration (hazard ratio 4.48) and non-aspiration pneumonia (hazard ratio 1.44).
- Study 4 was a prospective longitudinal study focusing on stroke patients admitted to a general hospital, investigating post-stroke dysphagia prevalence, related risk factors, and subsequent complications (22). Among the 395 stroke patients examined (184 female; mean age 73.2 years), a 45.06% prevalence of dysphagia was identified upon admission, with specific independently-associated risk factors including age, previous stroke history, stroke severity measured by the National Institute of Health Stroke Scale, and the volume of the lesion. Post-stroke dysphagia was independently linked to prolonged hospital stays, post-discharge institutionalization, diminished functional capacity, and notably higher mortality rates 3-month post-stroke. The study found that stroke severity and the patient's status before the stroke played more pivotal roles in dysphagia development than the exact location of the lesion.
- Study 5 was an open-label trial of 186 hospitalized elderly patients with dysphagia (20). It evaluated the Minimal-Massive Intervention for reducing nutritional and respiratory complications in this population. 62 with dysphagia (29 female; mean age 84.8 years) received the Minimal-Massive Intervention, while an equivalent number formed the control group matched by sex, age, functionality, comorbidities, and body mass index followed standard clinical practices. Assessments included geriatric, comorbidity, functionality, frailty, oral dysphagia, nutritional status, and oral health measures. The Minimal-Massive Intervention encompassed fluid and food texture modifications, caloric and protein supplementation, and oral health guidance during and after hospitalization. The Minimal-Massive Intervention group showed significant improvements in nutritional status and functionality, lower hospital readmissions and respiratory infections, and higher 6-month survival rates than the control group.

Statistical analysis

This study presents a comprehensive analysis of findings from five selected studies. Each study used univariate or multivariate analyses, calculating measures like odds ratios, incidence ratios, or hazard ratios to assess the associations between dysphagia and complications. Statistical methods included chi-square tests, Fisher exact tests, Mann–Whitney U tests, logistic regression, Cox regression, and survival analysis, with a consistent significance level set at $p < 0.05$. Our study synthesized these findings into a tabulated format, providing a consolidated overview of the associations between dysphagia and clinical outcomes, aiming to represent the statistical outcomes reported in the original studies comprehensively.

Results

Five studies conducted between 2001 and 2014 were included in data compilation, comprising a cumulative cohort of 3,328 patients with dysphagia uniformly managed in the same institution. Our compilation in Table 1 summarized and presented the findings from these studies, revealing substantial associations between dysphagia and a range of clinical complications among older individuals and patients with neurological conditions.

The data synthesis from these studies highlighted significant relationships between dysphagia and various clinical outcomes. For instance, older individuals with dysphagia and pneumonia exhibited a significant increase in 1-month mortality (odds ratio: 3.28, 95% CI: 1.13–9.50, $p < 0.05$), emphasizing the impact of pneumonia on short-term survival rates. Similarly, there was a substantial rise in 1-year mortality among this population (odds ratio: 3.42, 95% CI: 1.64–7.11, $p < 0.05$), underscoring the long-term consequences of pneumonia.

Additionally, our compilation emphasized the association between dysphagia and subsequent risks, such as malnutrition among older individuals from the community (odds ratio: 2.72, 95% CI: 1.25–5.95, $p < 0.05$), highlighting the importance of early nutritional evaluation and intervention.

Moreover, individuals with impaired safety of swallow, particularly from the community, demonstrated increased susceptibility to lower respiratory tract infections with notable odds ratios ranging from 2.39 to 2.55 (95% CI: 1.07–5.34 to 1.07–6.09, $p < 0.05$), emphasizing the association between swallowing difficulties and lower respiratory tract infections.

We observed influences on readmissions among older patients, especially from acute geriatric units, suggesting heightened odds for readmission, particularly for causes related to lower respiratory tract infections and other unrelated causes (odds ratios ranging from 1.37 to 5.97, 95% CI: 1.02–1.84 to 2.73–9.43, $p < 0.05$).

Additionally, post-stroke hospitalized patients with dysphagia exhibited a wide array of complications, including neurological complications, respiratory infections, urinary infections, and prolonged hospital stays. Odds ratios ranged from 2.11 to 27.34 (95% CI: 1.58–7.25 to 3.63–205.9, $p < 0.001$), signifying substantial associations between post-stroke complications and adverse outcomes.

Discussion

Dysphagia is a complex condition that significantly impacts the health of diverse patient groups. Our review offers a comprehensive synthesis and examination of data compiled from five previously published studies conducted at Mataro Hospital. This meticulous examination aimed to consolidate and synthesize findings from these investigations, offering a robust evaluation of dysphagia and its associated complications in older and post-stroke patients. The examination of data from these studies highlighted significant patterns and relationships, enriching our comprehension of the substantial impact of dysphagia within diverse clinical contexts among these patient phenotypes.

Our review reaffirms insights from previous studies, emphasizing the increased risks associated with dysphagia in various patient groups. These conclusions echo documented associations between dysphagia and increased risks, consolidating existing knowledge within diverse patient cohorts. The rise in mortality, consistent with some prior studies, underscores the need for targeted interventions for dysphagia in pneumonia cases. The heightened risks of aspiration pneumonia, malnutrition, hospital readmissions, and mortality rates among individuals with dysphagia highlight the urgent need for increased awareness and targeted interventions. Our findings show a profound correlation between dysphagia and aspiration pneumonia, particularly among older individuals, where pneumonia onset significantly escalates mortality rates at both 1 month and 1 year. Aspiration pneumonia arises from compromised swallowing safety, declining immunity and frailty due to aging, and inadequate oral hygiene, fostering bacterial colonization in the respiratory tract (24). Older individuals with swallowing difficulty often exhibit compromised oral health, heightened colonization of respiratory pathogens, and an increased susceptibility to lower respiratory infections. Recent research, notably highlighted in the Japanese Respiratory Society Guidelines, emphasizes the need for thorough investigation and management strategies despite the ambiguous diagnostic criteria for aspiration pneumonia (24). Current studies outline common clinical parameters for diagnosing aspiration pneumonia, focusing on symptoms, inflammatory markers, and specific chest imaging patterns associated with aspiration. However, distinguishing aspiration pneumonia from non-aspiration pneumonia presents challenges, prompting a shift toward a comprehensive approach that prioritizes evaluating swallowing function prospectively, identifying causal factors, and exploring alternative diagnoses or dysphagia-related causes in the older population experiencing pneumonia. Our study underscores the urgency of early recognition and management of dysphagia to mitigate the significant risk of aspiration pneumonia, a crucial step in enhancing survival rates among this vulnerable demographic. A recent scoping review aimed to identify clinical competencies for managing aspiration pneumonia in older adults (25). Ninety-nine studies were analyzed, resulting in a refined list of 12 competencies covering diagnosis, treatment, support, and interdisciplinary collaboration, emphasizing a "Diagnose, Treat and SUPPORT" approach. These competencies urge healthcare professionals to collaborate, address unmet needs, and enhance patient care, particularly focusing on supportive care aspects.

Our review revealed a significant association between dysphagia and subsequent malnutrition risk in older hospitalized patients, underscoring the need for prompt evaluation and intervention.

TABLE 1 Summary of findings from five studies on dysphagia (2001–2014): this table compiles findings from five studies conducted at Mataro Hospital, all focusing on dysphagia.

Complications	Phenotype	ODDS/IR/HR	*p* value	Analysis	Reference
1-month mortality	Older with pneumonia	3.28 (1.13–9.50) (OR)	Significant	Univariate	Cabre et al. (21)
1-year mortality	Older with pneumonia	3.42 (1.64–7.11) (OR)	-	Univariate	Cabre et al. (21)
MN and risk of MN	Older from the community	2.72 (1.25–5.95) (ODDS)	0.010	Univariate	Serra-Prat et al. (23)
LRTI	Older from the community (with ISS)	2.39 (1.07–5.34) (OR)	0.030	Univariate	Serra-Prat et al. (23)
MN and risk of MN	Older from the community (with IES)	2.31 (0.96–5.57) (OR)	0.062	Multivariate (adjusted)	Serra-Prat et al. (23)
LRTI	Older from the community (with ISS)	2.55 (1.07–6.09) (OR)	0.035	Multivariate (adjusted)	Serra-Prat et al. (23)
Pneumonia readmissions	Older patients from AGU	1.82 (1.41–2.36) (IR)	-	Univariate	Cabre et al. (7)
Non-aspiration pneumonia readmissions	Older patients from AGU	1.37 (1.02–1.84) (IR)	-	Univariate	Cabre et al. (7)
Aspiration pneumonia readmissions	Older patients from AGU	5.07 (2.73–9.43) (IR)	-	Univariate	Cabre et al. (7)
Bronchoaspiration readmissions	Older patients from AGU	4.36 (2.91–6.52) (IR)	-	Univariate	Cabre et al. (7)
Pneumonia readmissions	Older patients from AGU	1.60 (1.15–2.20) (HR)	0.005	Multivariate	Cabre et al. (7)
Non-aspiration pneumonia readmissions	Older patients from AGU	1.44 (1.02–2.03) (HR)	0.037	Multivariate	Cabre et al. (7)
Aspiration pneumonia readmissions	Older patients from AGU	4.48 (2.01–10.0) (HR)	<0.001	Multivariate	Cabre et al. (7)
Bronchoaspiration readmissions	Older patients from AGU	3.02 (1.73–5.27) (HR)	<0.001	Multivariate	Cabre et al. (7)
Mortality	Older patients from AGU	1.82 (1.62–2.04) (HR)	<0.001	Multivariate	Cabre et al. (7)
Neurological complications	Post-stroke hospitalized	3.38 (1.58–7.25) (OR)	<0.001	Univariate	Rofes et al. (22)
Respiratory infections	Post-stroke hospitalized	9.54 (2.80–32.55) (OR)	<0.001	Univariate	Rofes et al. (22)
Urinary infections	Post-stroke hospitalized	7.77 (1.72–35.2) (OR)	<0.001	Univariate	Rofes et al. (22)
Hospital stay	Post-stroke hospitalized	2.11 (beta coefficient)	0.049	Univariate	Rofes et al. (22)
Mortality	Post-stroke hospitalized	27.34 (3.63–205.9) (OR)	<0.001	Univariate	Rofes et al. (22)
Respiratory infections	3-month post-stroke	4.87 (2.25–10.54) (OR)	<0.001	Univariate	Rofes et al. (22)
Mortality	3-month post-stroke	17.46 (5.39–56.51) (OR)	<0.001	Univariate	Rofes et al. (22)
Respiratory infections	12-month post-stroke	2.28 (1.35–3.85) (OR)	0.003	Univariate	Rofes et al. (22)
Mortality	12-month post-stroke	11.40 (5.19–25.04) (OR)	<0.001	Univariate	Rofes et al. (22)
Hospital stay	Post-stroke hospitalized	0.938 (beta coefficient)	0.049	Multivariate (adjusted)	Rofes et al. (22)
Mortality	3-month post-stroke	6.90 (1.57–30.34) (OR)	0.011	Multivariate (adjusted)	Rofes et al. (22)
General readmissions	Older hospitalized	2.78 (150–5.15) (IR)	0.001	Univariate	Martín et al. (20)
LRTI readmissions	Older hospitalized	5.97 (1.45–24.63) (IR)	0.002	Univariate	Martín et al. (20)
Readmissions for other causes (no LRTI/pneumonia)	Older hospitalized	2.79 (1.21–6.44) (IR)	0.011	Univariate	Martín et al. (20)

Key variables include comorbidities (Charlson Comorbidity Index), functional abilities (Barthel Index), and nutritional status (MNA short-form). The studies used uniform statistical methods to calculate measures such as Odds ratio (OR), Incidence ratio (IR), and Hazard ratio (HR), assessing the associations between dysphagia and its complications. All studies consistently set the significance level at $p < 0.05$. MN, Malnutrition; LRTI, Lower respiratory tract infection; ISS, Impaired safety of swallow; IES, Impaired efficacy of swallow; AGU, Acute geriatric unit; OR, Odds ratio; IR, Incidence ratio; and HR, Hazard ratio.

Dysphagia can result in malnutrition, leading to deficiencies in crucial nutrients like proteins, calories, and vitamins due to swallowing difficulties (23, 26, 27). Additionally, reduced fluid intake can cause dehydration, particularly hypovolemic dehydration, leading to health complications. The triple adaptation concept offers a comprehensive strategy to tackle these issues (28). It primarily involves adapting food texture and fluid viscosity to ensure safer swallowing practices. Secondly, it customizes caloric, protein, and hydration intake to target individual nutritional deficiencies. Lastly, it enhances sensory attributes to encourage adherence to dietary guidelines. The practical application of triple adaptation resulted in 296 diverse recipes across 16 weekly menus based on Mediterranean cuisine principles. These recipes cater to various textures, viscosities, nutritional needs, and seasonal variations, and enhance the organoleptic quality of the dishes (28). This diet adaptation, integrating scientific insights into practical dietary interventions, allows for the management of both dysphagia complexities and prevalent nutritional deficiencies, improving compliance and clinical outcomes.

Our findings confirm a significant correlation between dysphagia and hospital readmissions due to clinical complications, primarily respiratory infections. Specifically, patients with poorly managed dysphagia have a 1.82 times higher risk of pneumonia readmission and a 5.07 times higher risk of aspiration pneumonia readmission. While our study did not directly investigate economic implications, it highlights the link between dysphagia and these complex clinical complications, indicating potential healthcare cost implications. Previous studies have stressed the financial burden of managing such complications, highlighting the extensive impact of untreated or poorly managed dysphagia on healthcare costs. This correlation emphasizes the importance of early dysphagia identification and effective management in reducing these complications. Proactive dysphagia management could potentially prevent these challenging clinical outcomes, thereby enhancing patient care and reducing healthcare resource strain.

The lack of dysphagia awareness and knowledge among caregivers, including general practitioners, is clear in our studies and wider literature (14–18). This knowledge gap presents a significant challenge in recognizing and managing dysphagia. To address this, we have implemented an innovative solution that uses AI to prospectively assess the risk of dysphagia in every patient admitted to our institution. We developed an expert system based on machine learning, using electronic health records of all hospitalized older patients during admission. This expert system calculates the risk of dysphagia, providing an accurate and systematic screening process. A recent study involving 2,809 older patients demonstrated the expert system's high predictive power, with a sensitivity of 0.940 and a positive predictive value of 0.834. The expert system efficiently screens all admitted patients in seconds, identifying those at greater risk of dysphagia and relaying this information directly to the clinician's workstation in real time. Currently in active use at our institution, this AI-driven expert system offers a significant advancement in the early recognition of dysphagia, enabling the implementation of tailored diagnostic and therapeutic strategies for each patient (19). In response to the absence of evidence-based treatment protocols for dysphagia, we have introduced Minimal-Massive Intervention, a promising shift in dysphagia management (20). This innovative method, designed for hospitalized dysphagia patients and based on aspiration pneumonia's pathophysiology, combines fluid modification, nutritional supplementation, and oral health recommendations (29). Our recent study demonstrated Minimal-Massive Intervention's effectiveness, showing significant improvements in nutritional status and functionality, reduced hospital readmissions and respiratory infections, and increased 6-month survival rates. These findings position Minimal-Massive Intervention as a promising, cost-effective strategy to mitigate dysphagia complications. By providing caregivers with the necessary knowledge and skills, including Minimal-Massive Intervention awareness, we can optimize dysphagia care and reduce associated risks and costs. Optimal Massive Intervention is an evolution from Minimal-Massive Intervention, offering a more comprehensive and personalized approach to dysphagia management. While Minimal-Massive Intervention focuses on three basic elements to prevent dysphagia complications in older hospitalized patients, Optimal Massive Intervention incorporates multiple components targeting improved swallowing function and enhanced quality of life across various dysphagia causes. Optimal Massive Intervention requires a multidisciplinary team, care coordination, evidence-based interventions, and continuous outcome monitoring, aligning with current best practice guidelines for dysphagia management. Optimal Massive Intervention is currently being evaluated in a clinical trial to assess its effectiveness and cost-effectiveness compared to standard care (30).

Our study, while insightful, has certain limitations. Its retrospective nature restricts our ability to establish causal relationships and control all confounding variables, despite our thorough analysis of Mataró Hospital's data. The study's focus on a single, internationally recognized European center may limit the applicability of our findings to wider populations. The data, sourced from a single hospital with high standards of assessment, clinical management, and follow-up, may not reflect the practices of other hospitals or geographical locations. While our findings offer valuable insights into the factors causing illness in older adults with dysphagia at Mataró Hospital, multi-center studies are needed to validate these results in other settings. Lastly, our study serves as a synthesis of our previous findings, contributing to its narrative review nature. This approach, while providing a comprehensive overview of our work on dysphagia complications, lacks the robustness of prospective experimental designs or systematic reviews.

After thorough problem identification and discussion, we have implemented a unique solution at our hospital that has shown promising results in addressing this issue. This is depicted in an algorithm (Figure 1) that showcases proactive universal screening, clinical diagnosis using volume-viscosity swallow test, and tailored management for dysphagia in older and post-stroke patients at Mataró hospital. Neurostimulation and transient receptor potential (TRP) stimulation are emerging as safe and effective interventions for dysphagia. Neurostimulation has been shown to enhance swallowing biomechanics, neurophysiology, induce cortical plasticity, and safety in people with dysphagia (31, 32). Electrical stimulation, including transcutaneous, neuromuscular, sensory, and pharyngeal, improves nerve or muscle function related to swallowing and has been shown to enhance swallowing safety, particularly in post-stroke patients with dysphagia (33–35). These techniques can enhance swallow response, reduce pharyngeal transit time, and lower penetration-aspiration scores. A low-intensity current treatment (25 mA, VitalStim device, FDA approved) endorsed by NICE guidelines (2018), is safe and prevents muscle damage and pain. However, it carries potential risks like muscle damage, infection, pain, and inconsistent effectiveness (36). High voltage stimulation may cause muscle damage if improperly administered (37). Non-invasive brain stimulation such as transcranial magnetic stimulation (TMS) and transcranial direct current stimulation (tDCS), can modulate the cortical activity related to swallowing by applying magnetic fields or direct currents to the scalp and can target specific brain regions involved in swallowing, such as the primary motor cortex, the premotor cortex, the supplementary motor area, and the insula (31, 38, 39). Non-invasive brain stimulation has been shown to enhance the swallowing function, reduce the aspiration risk, and increase the quality of life in people with dysphagia. Another promising treatment for dysphagia is the pharmacological stimulation of the TRP channels, which are expressed in the sensory nerves and epithelial cells of the oropharynx and larynx. TRP agonists, such as capsaicin, menthol, and piperine,

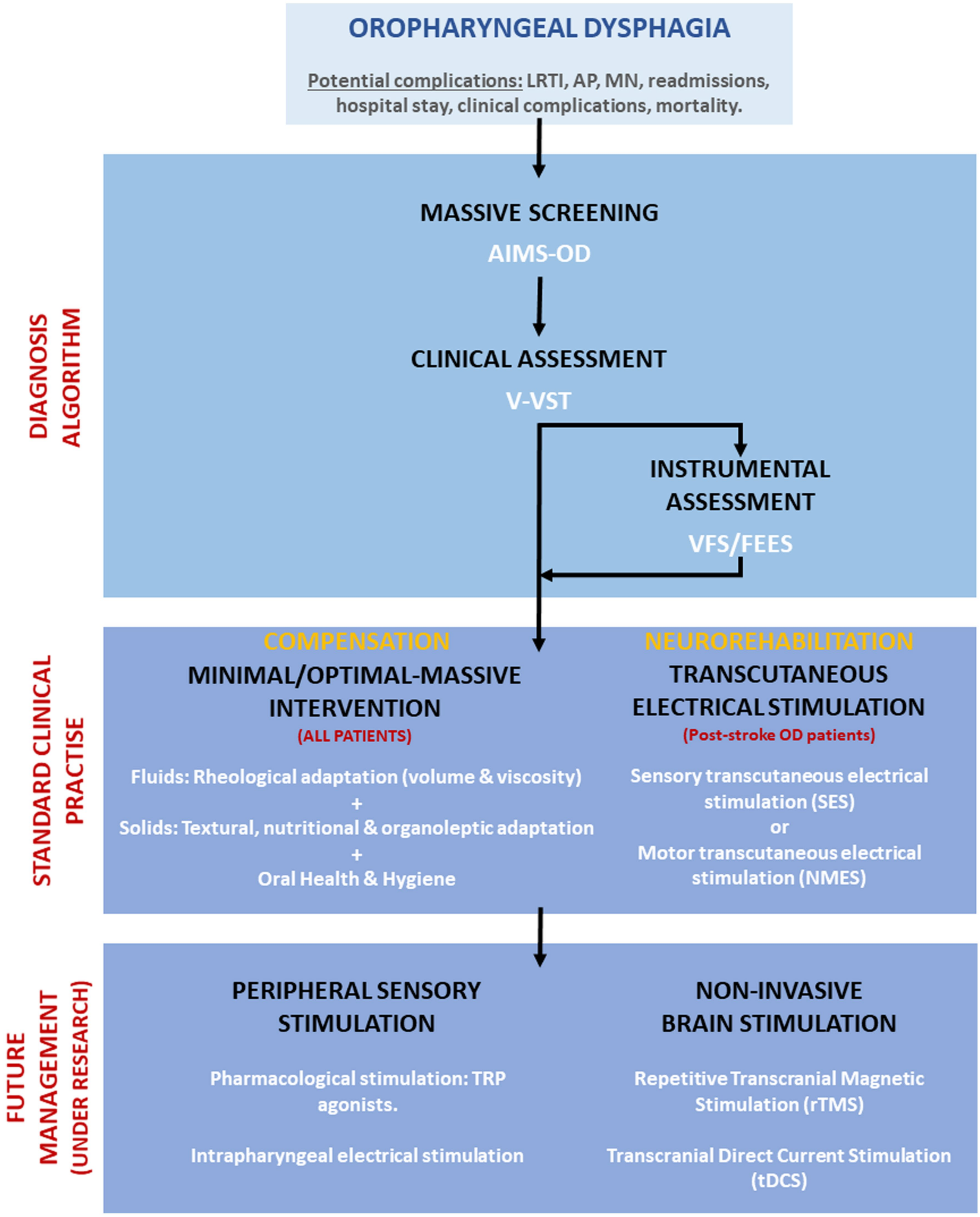

FIGURE 1
Algorithm for screening, diagnosis, and management of dysphagia at Hospital de Mataró. Future management treatments under research are also included. LRTI, Lower respiratory tract infections; AP, Aspiration pneumonia; MN, Malnutrition; AIMS-OD, Artificial intelligence massive screening for oropharyngeal dysphagia; V-VST, Volume-viscosity swallowing test; VFS, Videofluoroscopy; and FEES, Fiberoptic endoscopic evaluation of swallow.

can activate the TRPV1, TRPM8, and TRPV1/TRPA1 receptors, respectively, and modulate the sensory feedback and motor output of swallowing (31, 40). These treatments are well tolerated and do not cause major adverse events. Effectiveness of these treatments can vary significantly among individuals, influenced by factors like dysphagia's severity and cause, the patient's overall health, and treatment tolerance. Despite their potential demonstrated in clinical studies, further high-quality, large-scale randomized controlled trials are essential to confirm their efficacy and safety.

In conclusion, our review underscores the vital need for early recognition and intervention in dysphagia, particularly in older hospitalized patients and those with neurological complications. The studies we reviewed demonstrate that dysphagia is significantly linked with severe clinical complications like aspiration pneumonia in various phenotypes of older patients. The increased risks of aspiration pneumonia, malnutrition, hospital readmissions, and mortality rates among individuals with dysphagia highlight the urgent need for heightened awareness, systematic screening and diagnosis, and targeted interventions. It is essential to recognize dysphagia as a serious issue that warrants further attention and research.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

Ethical approval was not required for the study involving humans in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and the institutional requirements.

Author contributions

TK: Conceptualization, Writing – original draft, Writing – review & editing. PC: Conceptualization, Writing – original draft, Writing – review & editing. OO: Conceptualization, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Therapeutic singing-induced swallowing exercise for dysphagia in advanced-stage Parkinson's disease

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Background: With longer life spans and medical advancements, the rising number of patients with advanced-stage Parkinson's disease (PD) warrants attention. Current literature predominantly addresses dementia and fall management in these patients. However, exploring the impact of swallowing function on patients with advanced PD is crucial. Previous research has demonstrated notable enhancements in the quality of life related to voice for participants following a group singing-intervention program. To further elucidate the effect of individual singing-induced swallowing exercises, our study aimed to investigate the quantitative and qualitative effects of therapeutic singing on swallowing function in patients with advanced PD in comparison to a matched usual care control group. The hypothesis of this study is that therapeutic singing-induced swallowing exercises can assist to maintain swallowing function in patients with advanced PD.

Methods: This prospective matched control study compared the effects of a 6-week therapeutic singing-based swallowing intervention on swallowing function and quality of life in patients with advanced PD. The intervention group received individual sessions with a music therapist and conventional individual physical therapy. The control group received the same standard physical therapy for 6 weeks without music intervention. The primary outcome measure was Video Fluoroscopic Dysphagia Scale (VDS).

Results: The study revealed that the intervention group maintained swallowing function, whereas the control group experienced deterioration, indicating significant time-dependent changes in Penetration-Aspiration Scale (PAS), National Institutes of Health-Swallowing Safety Scale (NIH-SSS), and VDS. Analysis of PAS and NIH-SSS liquid food scores in both groups showed significant time effects. However, the intervention group exhibited no significant differences between the pre- and post-tests, indicating preservation of the swallowing function. VDS of liquid food indicated an interaction effect between time and group in the pharyngeal phase and total scores. The Swallowing-Quality of Life showed significant time-effect improvement in the intervention group.

Conclusion: Therapeutic singing exercises may help maintain swallowing function in advanced PD patients, potentially enhancing quality of life related to swallowing in those with advanced-stage diseases.

KEYWORDS

therapeutic singing, swallowing exercise, dysphagia, advanced stage, Parkinson's disease

1 Introduction

Advanced-stage Parkinson's disease (PD), defined as stages 4 and 5 on the Hoehn and Yahr scale, (1) is characterized by the deterioration of key functions including ambulatory dysfunction, worsening imbalance, and swallowing impairment (2, 3). It also includes the progression of non-motor symptoms and medication-induced adverse effects accelerating the decline in the quality of life (4). Declining efficacy of medications results from the diminishing dopaminergic neurons in the substantia nigra and a progressively lower capacity to store and convert exogenous dopamine (5). Despite variations in the predominance and severity of clinical phenotypes in advanced-stage PD, disability in the later stages is dominated by levodopa resistance and end-of-dose failure (6, 7). Current literature presents few clinical studies with patients entering the late disease stage, (8) leading to a lack of consensus because of ambiguous clinical characteristics in advanced and late-stage PD and unfavorable prognosis as the disease progresses to death (6). While improved general healthcare has increased longevity and improved clinical management of PD, the prevalence of advanced stage PD is expected to accelerate in the future. Therefore, more intensive and individualized interventions are required to address the complexity of the disease (9, 10).

Among the symptoms evident in the progression of PD, dysphagia is present in every stage of the disease; however, it becomes predominant in the advanced stages (11). PD symptoms lead to abnormal muscle movements causing oral and oropharyngeal dysfunction, (12, 13) and dysphagia often worsens with disease progression (14, 15). Thus, dysphagia emerges as a major cause of mortality and morbidity due to serious complications associated with dehydration, malnutrition and aspiration pneumonia (16).

Few therapeutic options, including levodopa optimization, are available for patients with advanced PD (17). However, dysphagia and dyskinesia are poorly controlled by existing drugs (18). Therefore, rehabilitative therapies are crucial to slow disease progression. Therapeutic strategies involving expiratory muscle strength training or electrical stimulation have shown improvements in degenerative function (coordination, speed, and volume), quality of life, and social relationships in individuals with PD (19). Previous literatures have highlighted the clinical assessment and therapeutic management of PD patients, often focusing on falls, postural instability, urinary dysfunction, freezing, bradykinesia, dysarthria, choking, dementia, psychosis, excessive daytime sleepiness, apathy, depression, and anxiety (20, 21). However, less interest has been channeled toward managing health-related quality of life in patients with advanced PD who are more likely to experience lifelong swallowing disabilities. Other studies have reported that the severity of dysphagia has a negative impact on an individual's quality of life (22). However, interventions targeting dysphagia have shown mixed effects on quality of life, with some articles reporting improvements that are not consistent across all interventions (23). For instance, enteral tube feeding, which is beneficial for maintaining physical health, also has drawbacks, as it leads to feelings of isolation (24, 25). Similarly, texture-modified food had both positive and negative effects on quality of life, as the appearance of such foods made individuals feel self-conscious and excluded from others (26). Prioritizing the treatment of non-motor complications, including dysphagia, is thus essential.

As a non-pharmacological intervention approach, a positive impact of a singing-integrated intervention on swallowing function in individuals with PD has been reported (27–29). Singing as a musically coordinated sensorimotor activity has been identified as a promising intervention considering its involvement in breathing, vocalization, and swallowing based on anatomical and neural correlation (30, 31). Singing requires increased control of respiratory, articulatory, and vocal organs (32, 33), and the process of singing intervention in our study has been modified to promote the laryngeal and articulatory movements that are involved in swallowing (34).

Significant improvements in swallowing function have been reported in previous studies after the completion of therapeutic singing-induced swallowing exercises in individuals with PD or head and neck cancer (27–29). Considering the additional benefits of singing such as emotional support and quality of life (35). Therapeutic singing-induced swallowing exercise could provide a comprehensive approach for patients with advanced PD. The hypothesis of this study is that therapeutic singing-induced swallowing exercises can assist to maintain swallowing function in patients with advanced PD. This study therefore aimed to investigate the quantitative and qualitative effects of therapeutic singing on swallowing function in patients with advanced PD in comparison to a matched usual care control group on the primary outcome of VDS liquid food total score. We also aimed to determine the results on secondary measures of PAS, NIH-SSS, and SWAL-QOL.

2 Materials and methods

2.1 Study design and ethics

This prospective intervention study with a matched control group was approved by the Institutional Review Board of Yonsei University Health System (Approval No. 4–2012-0483, Approval No. 4–2022-0560) at Severance Hospital and was conducted in accordance with the Declaration of Helsinki. The study was registered under the Clinical Research Information Service (CRIS) with trial registration number KCT0008644. All participants provided written informed consent after the rights of the participants and the purpose, methods, benefits, and risks of the study were fully explained. The study was designed as a matched control study, in which one researcher aiming to align the control group with the intervention group based on sex, age, diagnostic presentation, and degree of severity.

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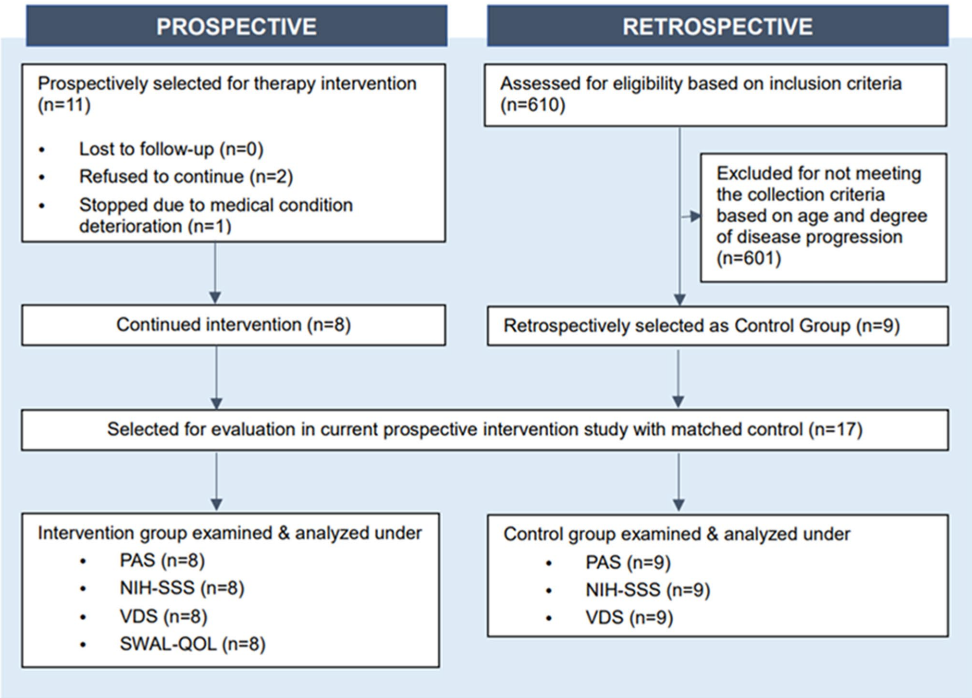

FIGURE 1
Patient disposition graph.

2.2 Participants and data source

Initially 11 patients were selected for therapy intervention; two of them autonomously declined to continue participation and one stopped the intervention due to deterioration of their medical condition (Figure 1). Therefore, eight patients with advanced PD completed the full course of therapeutic singing-induced swallowing exercises. All participants in the intervention group were recruited from the same rehabilitation hospital, where they provided legal consent to participate. The inclusion criteria for all participants of this study were (1) diagnosis of Parkinson's disease and Parkinsonism, (2) at or above stage 4 of the Hoehn and Yahr scale during the period of receiving PD-related medication and their usual antiparkinsonian treatment, (3) over 60 years of age, (4) no hearing impairment, and (5) no severe cognitive impairment. Matched control participants were selected from historical records. Six hundred ten patients admitted to the Severance Hospital, Seoul, South Korea, who received conventional therapy (excluding singing therapy) between 2017 and 2022 were reviewed and nine met the study inclusion criteria. Matching was performed by sex, age (±1 years), disease duration (±1 years), Hoehn and Yahr scale, and Modified Barthel Index score at initiation of admission, and swallowing function evaluation, as identified from Electronic Medical Record (Figure 1).

The target sample size was based on the results of another small music intervention study involving patients with PD ($n = 13$) with a similar dosage (36). To achieve an effect size of 0.60 with a fixed alpha error of 0.05 and power of 0.80 would require a sample size of 20.

2.3 Therapeutic singing-induced swallowing exercise

The intervention consisted of a 6-week program in which participants individually attended sessions with a music therapist twice a week for 30 min. This was in addition to the conventional therapy, which the control group also received. Conventional therapy was also administered twice a week for 30 min over 6 weeks. The control group followed traditional rehabilitation offers as scheduled. The components of this rehabilitation intervention included physical and occupational therapies.

The therapeutic singing-based swallowing intervention consisted of five parts as indicated in Supplementary Table 1: (1) respiratory muscle relaxation, (2) vocal warm-up, (3) vocal exercise for laryngeal elevation, and (4) modified singing. Step 1: Respiratory muscle relaxation involves stretching the neck muscles, lifting, and lowering the shoulders, and stretching the arms while participants maintain trunk balance. All movements in this step were accompanied by live electronic piano playing by a certified music therapist, and the dynamics of the music were adjusted to the participant's movement. Step 2: Vocal warm-up involves vocalizing single vowels, holding the breath for seconds, humming, and gliding a sound. The purpose of this stage is vocal fold relaxation, breathing control, and laryngeal movement. Step 3: Vocal exercises for laryngeal elevation consisted of singing two different notes from lower to higher pitches in a sequence with chord progression. Step 4: Modified singing focused on respiration (e.g., breathing control). Certified music therapists performed all intervention processes. The baseline measurement was assessed 1 week before the first session of the therapeutic singing

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intervention, and the post-test was assessed within 1 week after the intervention was terminated. Due to COVID19 our design was modified, requiring participants to strictly adhere to hospital policies of wearing masks during the physical preparation time. The participants also wore a transparent plastic face shield during therapy to prevent the spread of aerosols, including saliva. More time was needed to ensure that patients did not feel uncomfortable due to this additional personal protection equipment, and also that it did not hinder the treatment procedure.

The focus of our study on dysphagia rehabilitation centered on actions that involve the opening of the jaw. This emphasis stems from the understanding that the muscles responsible for hyolaryngeal elevation, corresponding to those involved in jaw-opening, play a crucial role in enhancing laryngeal elevation, similar to the effects of traditional swallowing exercises. Additionally, in steps 2 and 3 of our therapeutic singing intervention, patients are required to hum and glide across various pitch ranges, necessitating the precise control of their laryngeal muscles (Supplementary Table 1). A related source supports this by noting that "effortful pitch glide" induces notably greater excursions in anterior hyoid, superior hyoid, hyolaryngeal approximation, laryngeal elevation, and lateral pharyngeal wall medialization compared to swallowing (37). When participants underwent the pharyngeal squeeze maneuver that involved a forceful sound of "ee," the contraction of the pharyngeal constrictors allows improvement of pharyngeal strength (37). These findings suggest that the effortful pitch glide could be an effective exercise targeting crucial swallowing muscles, particularly the long pharyngeal muscles responsible for elevating the larynx and shortening the pharynx during swallowing (37). Similarly, the maneuver of pharyngeal muscles involved in singing therapy allows for these muscles to be trained and thus gain strength (37). Consequently, the mechanism of new exercise introduced in the aforementioned study supports our study design as training the muscles involved in singing can also have similar effect to swallowing exercise as they belong to the same group of muscles.

2.4 Outcome measures

The primary outcome measure of this study was VDS liquid food total score and the secondary outcome measures were PAS, NIH-SSS, and SWAL-QOL. Primary and secondary outcome measures were conducted at pre and post time point of 6-week intervention of therapeutic singing-induced swallowing exercise. A Video Fluoroscopy Swallowing Study was used to evaluate swallowing function in participants with PD. They performed swallowing tasks while ingesting three types of barium (one liquid and two solid powders). Participants in all groups swallowed 5 mL of three types of solution mixed with barium sulfate according to the instructions of the tester at the start of evaluation (12% semisolid solution consisted of barium sulfate, yogurt powder 9 g, Baritop HD power 9 g and 150 mL of water. 6% semisolid mixture consisted of barium sulfate, yogurt powder 4.5 g, Baritop HD power 4.5 g and 150 mL of water) (38). The outcomes of the videofluoroscopy swallowing study were evaluated using the penetration-aspiration scale (PAS), National Institutes of Health-Swallowing Safety Scale (NIH-SSS), and the video fluoroscopic dysphagia scale (VDS).

The PAS quantifies the degree of penetration and aspiration observed during the Video Fluoroscopy Swallowing Study and consists of an eight-point scale that assesses the depth of bolus passage into the airway and the patient's response. A higher PAS score indicates more severe symptoms (39). The PAS was used to evaluate changes in swallowing function. The NIH-SSS assesses swallowing stability by evaluating food residue, laryngeal penetration, aspiration response, maximal esophageal entry, and multiple swallows based on videofluoroscopic swallowing study observations (40). A higher score indicate more severe dysphagia (40). The NIH-SSS was used solely in the intervention group to assess changes in the swallowing function.

The VDS is a 14-item scale that assesses the oropharyngeal function during swallowing (41). The items included lip closure, bolus formation, mastication, apraxia, tongue-to-palate contact, premature bolus loss, oral transit time, pharyngeal swallowing triggering, vallecular residue, laryngeal elevation, pyriform sinus residue, coating of the pharyngeal wall, pharyngeal transit time, and aspiration (41). The first seven items assess the oral phase, whereas the next seven items evaluate the pharyngeal phase (41). The VDS scores range from 0 to 100, with higher scores indicating poorer swallowing function (41).

The SWAL-QOL was used to assess the quality of life of patients with oropharyngeal swallowing disorders (42). It consists of 11 subcategories and 44 questions (42). Higher scores indicate a higher quality of life associated with swallowing impairment (42). Two separate licensed physiatrists and two separate researchers with expertise in the related field conducted the outcome measures, and inter-rater reliability was tested.

2.5 Statistical analysis

Participants with advanced PD who underwent therapeutic singing-induced swallowing exercises were retrospectively compared with matched controls. Two-way repeated measures ANOVA and paired t-tests were used to analyze changes in swallowing functions over 6 weeks for each group and compare differences ($p < 0.05$ was set as criterion for statistical significance).

3 Results

3.1 Participants

After evaluating their eligibility for the study, the participating patients were divided into the intervention ($N = 8$) and control ($N = 9$) groups (Figure 1). The mean age of the participants in the intervention was 71.88 years (range of 56–86) with a mean onset duration of motor symptoms of 8.86 years and onset duration of dysphagia of 2.76 years. The mean age of the participants in the control group was 65.44 years (range–53–86) with a mean onset duration of 3.00 years. The level of disability of all participants was assessed using the Hoehn and Yahr scale. There were no significant differences between the intervention and control groups in terms of sex ($p = 0.229$), age ($p = 0.277$), duration of motor symptom onset ($p = 0.923$), duration of dysphagia onset ($p = 0.960$), Hoehn and Yahr stage ($p = 0.819$), or Modified Barthel Index total score ($p = 0.847$) (Table 1). The Modified Barthel Index

TABLE 1 Clinical characteristics of participants.

Characteristics	Intervention group (n = 8)	Control group (n = 9)	p-value
Gender			
Male, N (%)	5 (62.50%)	3 (33.33%)	0.229
Female, N (%)	3 (37.50%)	6 (66.67%)	0.229
Age (years), (mean ± SEM)	71.88 ± 3.54	65.44 ± 4.09	0.277
Onset duration, years			
Motor Symptom	8.86 ± 2.04	8.89 ± 1.76	0.923
Dysphagia	2.76 ± 1.42	1.78 ± 0.55	0.960
Hoehn and Yahr stage, N (%)			
Stage 4	4 (50.00%)	4 (44.44%)	0.819
Stage 5	4 (50.00%)	5 (55.56%)	0.819
Modified Barthel Index			
Personal hygiene	3.38 ± 0.56	2.78 ± 0.68	0.619
Bathing self	1.75 ± 0.49	2.22 ± 0.68	0.619
Feeding	6.13 ± 1.09	5.00 ± 1.52	0.767
Toilet	4.38 ± 1.02	4.56 ± 1.37	0.921
Stair climbing	3.13 ± 1.08	4.78 ± 1.45	0.398
Dressing	6.13 ± 0.79	6.00 ± 1.19	0.960
Bowel control	7.63 ± 1.25	5.67 ± 1.51	0.363
Bladder control	6.63 ± 1.25	5.56 ± 1.63	0.763
Ambulation	7.25 ± 1.68	7.89 ± 1.93	0.729
Chair/bed transfer	9.38 ± 1.61	9.33 ± 2.07	0.802
Total score	55.38 ± 9.74	53.78 ± 13.32	0.847

Data are presented as mean ± SEM or n (%). MBI, Modified Barthel Index, score range of 0 to 100. FIM, Functional Independence Measure, score range of 18 to 126. Lower scores of Modified Barthel Index and Functional Independence Measure indicate higher degree of dependence on caregivers while carrying out activities of daily living.

score criteria also did not show any statistical difference between the two groups (Table 1).

3.2 Swallowing function

The PAS, NIH-SSS, and VDS scores were obtained during the videofluoroscopy swallowing study. Two independent scorers had assessed PAS, NIH-SSS, and VDS for the nine patients in the control group (during their follow-up earlier), and the same scorers evaluated the scores for the eight participants in the intervention group later. The inter-rater reliabilities of the PAS, NIH-SSS, and VDS demonstrated intra-class correlation coefficients (ICC) of 1.00, 0.90, and 0.99, respectively. When comparing changes in the PAS and NIH-SSS scores between the intervention and control groups, both scales exhibited a significant time-effect before and after treatment for liquid food scores ($F_{1,}$ = 6.480, p = 0.022; $F_{1,}$ = 4.745, p = 0.046) (Table 2). The results indicated that the swallowing function of the control group weakened, showing significant time-dependent changes in the PAS and NIH-SSS in the pre- and post-treatment assessments (p = 0.045, p = 0.009, respectively, by Paired T-test). However, the results showed no significant differences between the pre-test and post-test PAS and NIH-SSS liquid substance scores in the intervention group, indicating that the swallowing function of the intervention group was maintained (p = 0.173, p = 0.732 by Paired T-test) (Table 2).

The VDS of liquid food showed an interaction effect between time and group in the pharyngeal phase and the total scores (F = 9.425, p = 0.008; F = 5.859, p = 0.029) (Table 3). The VDS of liquid food also showed time effect in the pharyngeal phase and the total scores (F = 15.017, p = 0.001; F = 14.864, p = 0.002) (Table 3). The results showed significant decrease in function over time in the pharyngeal phase and total VDS scores of the control group (p < 0.001), indicating impaired swallowing function in the control group, especially in the pharyngeal phase (pharyngeal phase: p = 0.642; total score: p = 0.439 by Paired T-test). Conversely, the swallowing function of the intervention group was maintained, with no significant differences between the pre- and post-test pharyngeal phases and total VDS scores (Table 3). Each VDS parameter was also analyzed to identify specific phase parameters influenced by the intervention, which revealed no significant change in the oral phase. In the pharyngeal phase, the VDS scores of coating on the pharyngeal wall showed a time-effect (F = 5.674, p = 0.031), interaction effect (F = 5.674, p = 0.031), with no group effect (F = 1.471, p = 0.244), and the results indicated that there was a significant difference between pre- and post-test scores of coating on the pharyngeal wall in the control group (p = 0.035 by Paired T-test), whereas there was no difference in the intervention group (p = 0.351 by Paired T-test) (Table 4). Pharyngeal transit time showed a group effect (F = 4.706, p = 0.047), but did not show time effect (F = 2.017, p = 0.176) and interaction effect (F = 2.017, p = 0.176) (Table 4). However, this should be carefully interpreted because of the lack of statistical significance of the time-dependent changes in the intervention group. Prior to the commencement of treatment, there was no significant difference in the pharyngeal transit time scores between the control group and the intervention group (p = 0.169 by independent T-test). However, following 6 weeks of music therapy, a noteworthy delay in pharyngeal transit time was observed in the control group compared to the intervention group (p = 0.031 by independent T-test). The aspiration results indicated that the swallowing function in the control group was significantly weakened (p = 0.022 by Paired T-test), especially in the pharyngeal phase, whereas the swallowing function in the intervention group was maintained (Tables 3, 4).

SWAL-QOL was measured only in the intervention group. The largest observed difference was found in the symptom frequency between pre- and post-intervention, although this did not reach statistical significance. Additionally, a significant difference between the pre-test and post-test was observed for the total SWAL-QOL score (p = 0.005), including subcategories such as food selection (p = 0.015), fear (p = 0.020), mental health (p = 0.009), social functioning (p = 0.036), and fatigue (p = 0.015), as shown in Table 5.

4 Discussion

The present study examined the effects of therapeutic singing-induced swallowing exercises on the PAS, NIH-SSS, VDS, and SWAL-QOL in patients with advanced PD. This is the first study to explore the effects of singing-integrated intervention on swallowing

TABLE 2 Swallowing functions in Parkinson's disease patients with dysphagia after singing-induced swallowing exercises.

Variables	Intervention group		Control group		Time		Group		Time * Group	
	Pre	Post	Pre	Post	F	*p*	F	*p*	F	*p*
PAS										
12% semisolid food	1.88 ± 0.88	1.00 ± 0.00	2.33 ± 0.88	1.89 ± 0.65	0.870	0.366	0.871	0.366	0.093	0.765
6% semisolid food	2.00 ± 0.87	1.50 ± 0.33	2.44 ± 0.87	2.00 ± 0.65	0.743	0.402	0.301	0.591	0.003	0.960
Liquid food	2.63 ± 0.65	3.63 ± 1.02	3.56 ± 0.96	5.67 ± 0.88#	6.480	0.022*	1.793	0.201	0.827	0.378
NIH-SSS										
12% semisolid food	3.25 ± 0.31	3.13 ± 0.13	3.78 ± 0.43	3.78 ± 0.36	0.144	0.709	1.700	0.212	0.144	0.709
6% semisolid food	3.88 ± 0.30	3.75 ± 0.25	3.89 ± 0.39	4.00 ± 0.44	0.002	0.964	0.073	0.791	0.614	0.445
Liquid food	4.13 ± 0.35	4.25 ± 0.25	4.11 ± 0.39	5.00 ± 0.33##	4.745	0.046*	0.769	0.394	2.693	0.122

Note. Data are presented as mean ± SEM. PAS, Penetration-Aspiration Scale with score range of 1 to 8; NIH-SSS, NIH-Swallowing Safety Scale with score range of 0 to 42. Higher scores indicate worsening of symptoms for PAS and NIH-SSS. *$p < 0.05$ significant effect by two-way repeated-measures ANOVA. #$p < 0.05$, ##$p < 0.01$ significant difference by Paired *T*-test. Data are presented as mean ± SEM.

TABLE 3 Swallowing functions in Parkinson's disease patients with dysphagia after singing-induced swallowing exercises.

VDS	Intervention group		Control group		Time		Group		Time * Group	
	Pre	Post	Pre	Post	*F*	*p*	*F*	*p*	*F*	*p*
12% semisolid food										
Oral phase	11.13 ± 4.01	10.56 ± 3.60	14.00 ± 4.09	16.39 ± 4.42	0.813	0.381	0.586	0.456	2.123	0.166
Pharyngeal phase	20.44 ± 4.41	26.69 ± 4.11	20.72 ± 5.24	22.78 ± 4.00	3.685	0.074	0.091	0.767	0.940	0.348
Total score	31.56 ± 7.51	37.25 ± 6.98	34.72 ± 8.13	39.17 ± 7.89	4.039	0.063	0.057	0.814	0.061	0.809
6% semisolid food										
Oral phase	11.13 ± 4.01	12.00 ± 3.97	13.67 ± 4.21	15.28 ± 4.66	1.251	0.281	0.241	0.631	0.110	0.745
Pharyngeal phase	24.25 ± 4.72	26.88 ± 3.97	22.50 ± 4.87	28.11 ± 4.15#	9.077	0.009**	0.002	0.967	1.193	0.292
Total score	35.38 ± 7.64	38.88 ± 6.82	36.17 ± 7.72	43.39 ± 7.99#	7.999	0.013*	0.062	0.806	0.964	0.342
Liquid food										
Oral phase	7.88 ± 3.67	10.13 ± 3.99	13.00 ± 3.97	15.72 ± 4.54	3.644	0.076	0.903	0.357	0.033	0.859
Pharyngeal phase	27.31 ± 3.38	28.88 ± 4.41	22.67 ± 5.34	36.44 ± 4.22###	14.864	0.002**	0.059	0.811	9.425	0.008*
Total score	35.19 ± 5.90	39.00 ± 6.70	35.67 ± 7.55	52.17 ± 7.31###	15.017	0.001**	0.513	0.485	5.859	0.029*

Data are presented as mean ± SEM. VDS, Videofluoroscopic Dysphagia Scale, with total score range of 0 to 100. Higher scores indicate worsening of symptoms. *$p < 0.05$, **$p < 0.01$ significant effect by two-way repeated measures ANOVA. #$p < 0.05$, ###$p < 0.001$ significant difference by Paired *T*-test.

function in patients with advanced PD using a prospective intervention study with matched control group. The findings of this study indicated that minimal changes were observed in most parameters between the pre-test and post-test assessments in both the intervention and control groups. However, a clinically significant observation was made regarding the scores in the liquid food condition, serving as an indicator of a higher risk of aspiration. The intervention group exhibited stable scores in this condition, whereas the control group demonstrated a statistically significant decline in functional ability in both the PAS and NIH-SSS under liquid condition testing.

VDS is a comprehensive tool utilized in Video Fluoroscopic Swallowing Studies (VFSS) to assess swallowing function. It evaluates oral and pharyngeal phases, penetration/aspiration, residue, and abnormalities, providing a detailed understanding of dysphagia nature and severity. Compared to PAS and NIH-SS, VDS identifies swallowing issues in advanced PD patients due to its extensive parameters. It offers objective measurements, reducing subjectivity and enabling reliable comparisons. Standardized scoring ensures consistency across clinicians and settings, fostering communication and interpretation. Widely used in clinical and research settings, VDS predicts clinical outcomes like aspiration pneumonia risk and treatment response, aiding patient management decisions. It monitors swallowing changes over time for tailored interventions and improved outcomes.

When statistical analyses are conducted to compare different dysphagia scales, the significance of the results can vary based on several factors, including the sensitivity of the scales and the number of parameters they assess. Generally, scales that evaluate a broader range of parameters related to swallowing function may yield more significant results in statistical analyses. In this case, VDS scores are more sensitive that NIH-SS, and thus have shown more significant changes between the comparing groups. Scales that assess multiple parameters of swallowing function, such as VDS, are inherently more sensitive to variations in swallowing function. This increased sensitivity allows for the detection of subtle differences or abnormalities that may not be captured by scales focusing on a narrower range of parameters, such as PAS.

TABLE 4 Swallowing functions in Parkinson's disease patients with dysphagia after singing-induced swallowing exercises.

VDS	Intervention group		Control group		Time		Group		Time * Group	
	Pre	Post	Pre	Post	*F*	*p*	*F*	*p*	*F*	*p*
Lip closure	0.25 ± 0.25	0.50 ± 0.33	0.00 ± 0.00	0.00 ± 0.00	1.134	0.304	2.306	0.150	1.134	0.304
Bolus formation	2.25 ± 0.94	2.63 ± 1.05	2.67 ± 0.93	3.00 ± 0.87	0.941	0.347	0.095	0.763	0.003	0.955
Mastication	2.50 ± 1.30	3.00 ± 1.46	3.56 ± 1.24	4.00 ± 1.15	0.941	0.347	0.344	0.566	0.003	0.955
Apraxia	0.00 ± 0.00	0.19 ± 0.19	0.17 ± 0.17	0.33 ± 0.33	2.008	0.177	0.311	0.585	0.007	0.935
Tongues to palate contact	0.63 ± 0.63	0.63 ± 0.63	2.78 ± 0.88	3.89 ± 1.39	2.017	0.176	4.261	0.057	2.017	0.176
Premature bolus loss	1.88 ± 0.74	2.44 ± 0.75	2.50 ± 0.79	2.83 ± 0.73	1.982	0.180	0.251	0.623	0.130	0.724
Oral transit time	0.38 ± 0.38	0.75 ± 0.49	1.33 ± 0.53	1.67 ± 0.53	2.008	0.177	2.090	0.169	0.007	0.935
Triggering of pharyngeal swallow	0.56 ± 0.56	0.56 ± 0.56	2.00 ± 0.79	2.50 ± 0.79	0.882	0.362	3.107	0.098	0.882	0.362
Vallecular residue	2.00 ± 0.00	2.25 ± 0.25	2.67 ± 0.33	3.11 ± 0.48	3.151	0.096	3.151	0.096	0.247	0.626
Laryngeal elevation	5.63 ± 1.65	5.63 ± 1.65	2.00 ± 1.32	5.00 ± 1.58	1.765	0.204	1.281	0.275	1.765	0.204
Pyriform sinus residue	4.50 ± 0.00	5.06 ± 0.56	6.00 ± 0.75	6.50 ± 0.79	2.008	0.177	3.211	0.093	0.007	0.935
Coating on the pharyngeal wall	7.88 ± 1.13	7.88 ± 1.13	4.00 ± 1.58	8.00 ± 1.00#	5.647	0.031*	1.471	0.244	5.647	0.031*
Pharyngeal transit time	0.00 ± 0.00	0.00 ± 0.00	1.33 ± 0.88	2.67 ± 1.05	2.017	0.176	4.706	0.047*	2.017	0.176
Aspiration	6.75 ± 1.36	7.50 ± 1.88	4.67 ± 1.67	8.67 ± 1.45#	4.486	0.051	0.054	0.819	2.100	0.168

Data are presented as mean ± SEM. VDS, Video fluoroscopic Dysphagia Scale, with total score range of 0 to 100. Higher scores indicate worsening of symptoms. *$p < 0.05$ significant effect by two-way repeated-measures ANOVA. #$p < 0.05$ significant difference by Paired *T*-test.

TABLE 5 SWAL-QOL scores before and after singing-induced swallowing exercises for intervention group (*N* = 8).

Parameter	Pre	Post	*t*	*p*
Burden	7.25 ± 1.48	7.38 ± 0.74	−0.196	0.850
Eating duration	6.13 ± 1.55	7.13 ± 1.55	−1.080	0.316
Eating desire	9.63 ± 2.06	9.63 ± 2.20	0.000	1.000
Symptom frequency	40.25 ± 5.44	49.88 ± 1 4.63	−0.2.064	0.078
Food selection	4.63 ± 1.76	6.63 ± 0.91	−3.191	0.015*
Communication	5.00 ± 2.50	6.00 ± 1.41	−1.366	0.214
Fear	9.13 ± 3.04	12.88 ± 2.10	−0.794	0.020*
Mental health	10.63 ± 2.72	15.25 ± 2.18	−0.3.572	0.009**
Social functioning	12.13 ± 2.69	15.63 ± 2.44	−2.593	0.036*
Fatigue	5.75 ± 2.37	8.00 ± 1.06	−3.211	0.015*
Sleep	4.13 ± 1.55	5.13 ± 1.80	−1.038	0.334
Total	114.63 ± 12.10	143.50 ± 17.92	−3.998	0.005**

Data are presented as mean ± SEM. SWAL-QOL, Swallowing Quality of life questionnaire, with total score range of 0 to 100. Lower scores indicate higher degree of impairment in the quality of life. *$p < 0.05$, **$p < 0.01$ significant effect by two-way repeated measures ANOVA.

Given the non-linear trajectory of symptom progression observed in patients with advanced PD (43), preservation of functional levels becomes clinically important. This significance stems from the recognition that advanced PD involves a multitude of pathological alterations within the neuromuscular structures associated with swallowing, leading to impairments across all stages of the swallowing process. Moreover, aging is associated with reduced muscle tone in the elderly population, resulting in reduced chewing and swallowing capabilities, rendering patients with PD more susceptible to dysphagia (44). Considering the severity of PD as indicated by Hoehn and Yahr stages 4 and 5 in the participants, singing interventions can be beneficial in delaying symptomatic regression.

The improvement in SWAL-QOL after 6 weeks of therapeutic singing-induced swallowing exercise showed that music experience positively influenced the participants' quality of life. Due to the retrospective nature of the study, the analysis of SWAL-QOL scores was only feasible within the intervention group. Upon examining the subcategories of the SWAL-QOL, improvements in psychosocial indicators were observed. The positive outcomes of music therapy in various functional domains of PD have been consistently reported in previous studies (45–47). Singing has been widely acknowledged for its effectiveness in enhancing emotional aspects and communication abilities, and the SWAL-QOL findings in this study support the results of previous studies (33). A study by Tamplin et al. (33) suggests that singing by nature allows patients to express and internalize their emotions better, especially when they lack the ability to communicate their feelings actively to caregivers and healthcare providers. This renowned study by Tamplin et al. (33) has proved the significant improvements in voice-related quality of life for Parkin Song participants in terms of acoustic measures of vocal loudness, with a perceived decrease in severity of voice problem, and less negative feelings about their voice problem in addition to mood enhancement and stress relief. Singing may lead to improved mood with its physiological and neurochemical effects. Singing demonstrated a positive association with diminished cortisol, beta-endorphin, and oxytocin levels, suggesting a potential modulatory impact on both mood states and components of the immune system in this preliminary investigation involving cancer patients (48). In this study, enhancements in mood were notably pronounced among individuals with lower mental wellbeing, revealing substantial positive associations between mood changes and baseline anxiety and depression levels, alongside noteworthy negative correlations with baseline levels of wellbeing and social resilience (48). The experience of music, an intangible stimulus, has the potential to elicit sensations of joy and a desire for more, akin to the gratification associated with concrete rewards linked to the dopaminergic system in the striatum (49). This implies that singing allows individuals to express and convey their emotions through music and lyrics. Singing-induced swallowing exercise adopted in our study thus can provide a healthy outlet for emotional expression and can be particularly therapeutic for patients with advanced PD dealing with stress, depression, or other challenging emotions. In addition, engaging in singing can serve as a distraction from negative thoughts and worries. Focusing on the act of singing and the enjoyment of music can redirect attention away from stressors that these patients with advanced PD may be more sensitive towards.

A similar study by Baker et al. (50) elucidated that engaging in recreational choir singing emerges as a therapeutically relevant intervention with clinical implications for alleviating depressive symptoms in individuals with dementia within the care home setting in Australia. Although this study was designed with singing in a group, the effect of singing itself was proven to alleviate depressive mood among patients with dementia which also involve cognitive decline like advanced PD. Successful participation in singing, whether individually like in our study design or in a group, can boost self-esteem and confidence. This suggests that accomplishing musical goals and receiving positive feedback from the therapists contribute to a sense of achievement for the patients with advanced PD and similar neurodegenerative diseases.

Another study by Tamplin et al. (51) targeting people with spinal cord injury patients reported that sensations of enjoyment were notably elevated following sessions of music therapy, reinforcing earlier discoveries indicating that group singing fosters a sense of happiness, positive mood, joy, elation, and a heightened feeling. The results from their study supports the results of our study including parameters such as "fear," "mental health" and "social functioning" in SWAL-QOL showing significant improvement. It can be inferred that participating in an intervention involving music has facilitated the experience of psychosocial support, fostering emotional bonds and promoting mental health status in participants. The greatest improvement was observed in decrease of symptom frequency in the intervention group, which includes severity of drooling and presence of excessive saliva or thick phlegm (52), although this was not statistically significant. Similar results have been reported in previous studies that used expiratory muscle strength training or speech-language therapy (53, 54).

Swallowing is a complex neural representation, involving multiple regions such as the caudal sensorimotor and lateral premotor cortex, insula, temporopolar cortex, amygdala, brainstem, and cerebellum (55). Neural activation for singing commonly occurs in the inferior precentral and postcentral gyrus, superior temporal gyrus and superior temporal gyrus, and superior temporal sulcus bilaterally, as these activated areas indicated a large shared network for motor preparation associated with vocalization, execution, and sensory feedback/control for vocal production. (33, 56). These neural processes have far-reaching implications, affecting various body systems involved in the act of swallowing. Thus, it might be interpreted that functional improvement in swallowing can promote patients' cognitive ability (53, 54). Patients with advanced PD can experience cognitive-linguistic impairments, poor self-perception of speech and neuropsychiatric symptoms including depression and anxiety (33). This is relatable to our study as it stems from the fundamental idea of singing requiring coordination of various cognitive processes such as memory, attention, and language. Engaging in these cognitive activities can have positive effects on overall cognitive function and mental well-being of patients with advanced PD.

The act of singing also requires patients to sustain their phonation, articulation and improve their respiratory support, as elucidated by previous studies of Tamplin et al. (33). This particular study by Tamplin et al. (33) emphasized on the improvement in speech functions related to articulation and breathing control, which also shares a similar focus to our study in mitigating and potentially reversing the neurodegenerative nature of the disease as it is crucial to maintain the use of swallowing muscles that are also used in singing. This can be achieved through consistent engagement in specific vocal, respiratory, and swallowing exercises tailored for individuals with advanced PD. Singing involves controlled breathing, which can have a calming effect on the nervous system. Deep, controlled breaths can reduce physiological responses to stress and promote relaxation, and training of muscles related to speech and swallowing. Singing, therefore, can serve as a promising therapeutic modality to improve speech related functions including swallowing as it enhances related muscles.

The mean levodopa dosage prescribed between the intervention group and control group in the current study did not demonstrate a statistically significant difference, suggesting that L-dopa had no known effect on the swallow function in the current study as a covariate. The applicability and efficacy of non-pharmacological treatments for speech impairment should be considered in the management of speech disorders in patients with PD. Moreover, a higher L-dopa dose does not always improve swallowing, and non-pharmacological interventions must be prioritized (57).

The advantage of the protocol used in this study is that it emphasizes the detailed mechanisms required for singing, such as breathing, vocalization, tongue base contraction, and articulation, while enabling the integrated control of complex functions through a single singing context. Patients can repeatedly perform this integrated training process as singing training is frequently used as a form of exercise training for patients who need to improve laryngeal function, and its effectiveness has been reported in several cases (38, 58). This study has important implications for the application of the effects of laryngeal muscle movements induced by therapeutic singing to assist in maintaining swallowing function.

Motor- and non-motor symptoms unresponsive to levodopa are the most reliable predictors of nursing home placement and mortality (59, 60). Clinicians have a strong tendency to focus their attention on conservative treatment of postural instability, falls, dementia and hallucinations in patients with advanced PD, the strongest independent predictors of institutionalization and death (59, 60). The clinical significance of this study arises from the novelty of assessing and treating dysphagia and the overall quality of life related to swallowing in patients with advanced PD, where the current literature mainly focuses on motor- and non-motor symptoms, excluding dysphagia and SWAL-QOL. The nature of patients with advanced PD and the heterogeneity in their symptoms also limits clinicians from conducting reliable clinical research; however, this study has presented treatment effects on patients with verified and holistic clinical characteristics.

This study serves as an important step in elucidating the impact of therapeutic singing-induced swallowing exercises on assistance of maintaining swallowing function in patients with advanced-stage PD at an individual level. The singing-induced swallowing protocol could be a potential treatment option for dysphagia in patients with PD. The clinical importance lies in providing an intervention that allows patients with advanced PD to participate and observe positive changes. One limitation of this study is that the SWAL-QOL was only assessed in the intervention group. For a more comprehensive assessment of swallowing function and quality of life in patients with advanced PD, the SWAL-QOL should be assessed in both the control and intervention groups. Other limitations include small sample and the absence of an active control group. In a future study, more patients need to be recruited as a randomized controlled trial.

Current medical practitioners may face difficulties in discerning significant changes in swallowing symptoms among patients with advanced-stage PD. As a result, we conducted the study using a 1:1 matching design and confirmed the effect of the intervention protocol through comparison with a control group. These experiences will provide researchers with suggestions for the importance of and insights into various interventions and measures. Further research should be conducted to investigate the long-term effects of therapy and to examine the effects of therapy on the quality of life of patients with advanced PD. This may involve multiple sessions of singing intervention over a longer period, providing better insight for clinicians to determine the ideal number of sessions and the degree of singing intervention depending on the progression of the disease and the type of neurodegenerative disease.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Institutional Review Board of Yonsei University Health System and registered under Clinical Research Information Service. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

MY: Conceptualization, Data curation, Methodology, Validation, Writing – original draft, Writing – review & editing. JH: Conceptualization, Investigation, Project administration, Validation, Visualization, Writing – original draft, Writing – review & editing. HL: Conceptualization, Data curation, Formal analysis, Investigation, Validation, Visualization, Writing – review & editing. SK: Conceptualization, Data curation, Investigation, Supervision, Writing – review & editing. S-RC: Conceptualization, Investigation, Project administration, Supervision, Validation, Visualization, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fneur.2024.1323703/full#supplementary-material

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Clinical study on the efficacy of postural control combined with electroacupuncture in treating dysphagia after stroke

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Objective: To evaluate the clinical effectiveness of combining postural control with electroacupuncture in the treatment of dysphagia following stroke, with the goal of establishing a solid clinical foundation for this therapeutic approach and investigating potential mechanisms to stimulate additional research and progress in post-stroke dysphagia management.

Methods: 138 patients who met the diagnostic and inclusion criteria were enrolled and divided into control group and observation group. Both groups received conventional rehabilitation training. Additionally, the control group received swallowing training and diet optimize, while the observation group received swallowing training, diet optimize, posture control as well as electroacupuncture therapy. After four weeks, swallowing function was assessed and compared between the two groups using the Standardized Swallowing Assessment (SSA) score and water swallowing test (WST).

Results: Patients who underwent postural control therapy in combination with electroacupuncture demonstrated significantly higher treatment efficacy compared to the control group. Notably, The SSA score and WST score in both groups decreased significantly, and the observation group showed more improvements in aspiration compared to the control group.

Conclusion: The integration of posture control, electroacupuncture, and conventional rehabilitation training can effectively lower the degree of post-stroke swallowing disorders, restore swallowing function, and significantly reduce the occurrence of complications such as aspiration, fever, and nutritional disorders. Moreover, this approach significantly improves the quality of life of patients and is more effective than conventional rehabilitation training in treating post-stroke swallowing disorders.

Clinical trial registration: https://www.chictr.org.cn/, Identifier ChiCTR2300075870.

KEYWORDS

stroke, rehabilitation, dysphagia, postural control, electroacupuncture

Introduction

Stroke frequently results in a range of functional impairments encompassing movement, sensation, swallowing, speech and language, cognition, mood, excretion, and cardiopulmonary functions (1–3). Post-stroke dysphagia (PSD) represents a prevalent complication, affecting an estimated 28% to 67% of stroke patients (4). Moreover, individuals with dysphagia following stroke often endure persistent swallowing difficulties, persisting up to six months post-onset (5). Neuromuscular Electrical stimulation (NMES), Pharyngeal electrical stimulation (PES) and modified Pharyngeal electrical stimulation (mPES), also have been reported as an effective treatment (6, 7). Nevertheless, the exact mechanism of NMES, PES and mPES is unclear, and there has been little consensus on optimal electrode placement as well as proper frequency and intensity for stimulation (8).

Acupuncture, rooted in the meridian theory, entails the targeted stimulation of specific acupuncture points to harmonize meridians and address ailments in a minimally invasive and relatively painless manner, rendering it readily acceptable to patients. Postural control for dysphagia post-stroke, derived from the Bobath technique, provides a neurodevelopmental framework for assessing and managing positional, motor, and functional deficits stemming from central nervous system injury (9). This approach prioritizes rectifying aberrant postures, fostering the restoration and enhancement of normal postural alignment, and ultimately facilitating nerve function improvement.

This study aimed to assess the efficacy of posture control combined with electroacupuncture targeting specific acupoints including Xiaguan (ST7), Jiache (ST6), Chengjiang (CV24), Lianquan (CV23), Jinjin (EX-HN12), Fengchi (GB20), and Yifeng (TE17) for the treatment of dysphagia post-stroke (10). The objective was to provide a scientific and objective evaluation of the treatment outcomes and investigate the underlying mechanisms. This innovative and efficacious rehabilitation approach offers promise in improving the management of dysphagia following stroke, reducing the risk of complications such as pneumonia, and ultimately enhancing patient survival rates.

Materials and methods

Patients

This study enrolled a total of 138 patients admitted to The Third Clinical Medical College of the Three Gorges University, Gezhouba Central Hospital of Sinopharm. These patients met both the diagnostic and inclusion criteria and were randomly assigned to either the observation group ($n = 69$) or the control group ($n = 69$). The group assignments followed the principles of randomized controlled trial (RCT) design. The study participants were individuals who experienced dysphagia during the recovery period after stroke. A few participants dropped out of the randomized clinical trial for various reasons. 5 participants in the observation group withdrew due to cerebral hemorrhage, while in the control group, 4 participants were lost to follow-up due to exacerbated pulmonary infection, and 2 participants were excluded from the study due to gastrointestinal bleeding during treatment. Ultimately, the observation group comprised 65 patients, and the control group comprised 62 participants.

Inclusion and exclusion criteria

The inclusion criteria utilized for participant selection in this study were as follows: (1) patients meeting the diagnostic criteria for stroke, as defined by both Traditional Chinese Medicine (TCM) (aligned with the "Scoring Standards for Stroke Diagnosis" issued by the Encephalopathy Collaborative Group of the State Administration of Traditional Chinese Medicine) and Western medicine (diagnostic criteria for cerebral infarction or cerebral hemorrhage in the "Key Points for the Diagnosis of Various Major Cerebrovascular Diseases in China 2019"). Additionally, the duration of their illness should fall within the range of 15 days to 6 months; (2) patients diagnosed with swallowing dysfunction based on the water swallowing test; (3) swallowing dysfunction confirmed through swallowing contrast examination; (4) absence of any other neurological diseases; and (5) patients with stable vital signs who were willing to cooperate with therapists for rehabilitation training and examination.

Exclusion criteria were defined as follows: (1) patients with concomitant conditions such as oropharyngeal lesions (e.g., pharyngitis, pharyngeal tumors, posterior pharyngeal wall abscesses), esophageal lesions (e.g., esophagitis, esophageal cancer, cardiac spasm), or psychiatric disorders (e.g., functional neurological disorders); (2) patients with ulcers or a predisposition to bleeding easily; (3) patients with organ failure that could significantly impact their overall health; and (4) individuals experiencing concurrent consciousness and mental disorders.

Based on the preliminary results of the pre-experiment, the treatment group exhibited an effective rate of 90%, while the control group showed an effective rate of 70%. Assuming a probability of type I error (α) of 0.05 and a power (1-β) of 90%, an estimation of the required sample size for the study was conducted. The sample size calculation was performed based on the primary efficacy outcome measures, following the design of the clinical trial study, using the formula outlined below.

$$n = \frac{p_1 \times (1 - p_1) + p_2 \times (1 - p_2)}{(p_1 - p_2)^2} \times \left(\mu_{\alpha/2} + \mu_\beta\right)^2$$

Where P1 = 90%, P2 = 70%, α = 0.05, β = 0.1 are substituted into the formula.

$$n = \frac{0.9 \times (1 - 0.9) + 0.5 \times (1 - 0.7)}{(0.9 - 0.7)^2} \times (1.96 + 1.28)^2$$

According to the above calculations, 63 cases are needed per group. Considering a 10% dropout rate, 69 cases should be enrolled per group. A total of 138 cases were reported in both groups.

Eligible participants

A total of 138 patients with dysphagia after stroke were enrolled in this study, and 127 patients were included according to the inclusion and exclusion criteria (Figure 1).

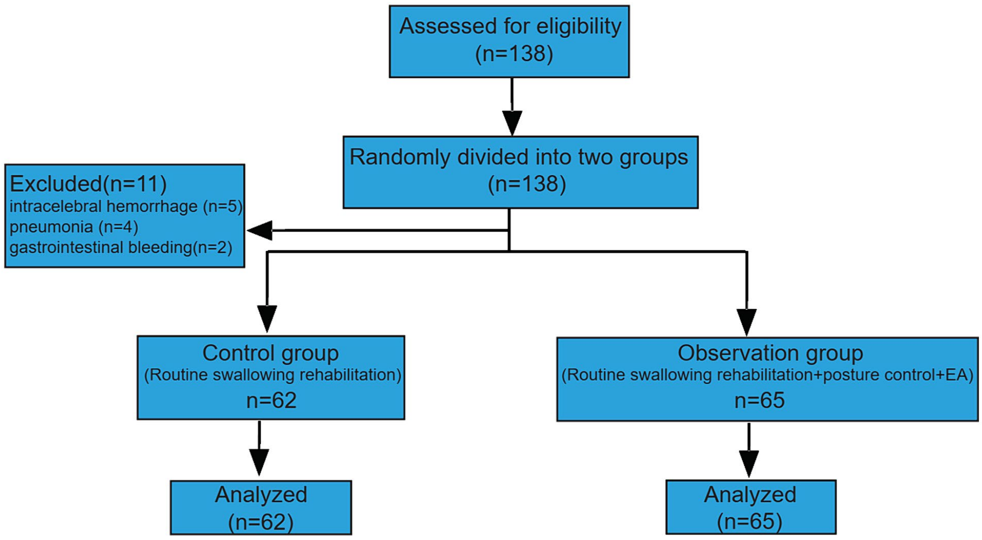

FIGURE 1
Schematic of the clinical trial.

Interventions and assessment of outcomes

Conventional swallowing rehabilitation typically encompasses several components: (1) enhancement of the physical properties of food; (2) adjustment of eating position; (3) functional rehabilitation training for oropharyngeal and facial muscles, including sensory stimulation, oral and facial muscle strength training, coordination training for swallowing muscle movement, and swallowing skill training; (4) electrical stimulation of the pharyngeal muscles, including Neuromuscular Electrical stimulation (NMES), modified pharyngeal electrical stimulation (mPES) (7) and Pharyngeal electrical stimulation (PES) (6, 8, 11); (5) application of cold and hot stimulation to stimulate the pharyngeal mucosa; (6) Specific training for laryngeal elevation, hyoid muscle function, cricopharyngeal muscle function, pharyngeal cavity power, vocal cord function, and airway protection (11–13).

In terms of acupuncture point selection and electroacupuncture treatment, this study adhered to the principle of proximal acupoint extraction and targeted strategic points such as Xiaguan (ST7), Jiache (ST6), Chengjiang (CV24), Lianquan (CV23), Jinjin (EX-HN12), Fengchi (GB20), and Yifeng (TE17). Patients were positioned either seated or supine, and standard disinfection of the acupuncture points was performed prior to using disposable acupuncture needles measuring 0.25 mm × 40 mm for electroacupuncture application using conventional acupuncture techniques. To ensure effective stimulation of the acupoints, Han's Acupoint Nerve Stimulator (LH202, Huawei Co. Ltd., Beijing, China) was employed, delivering a continuous wave with a current intensity of 1 mA and a frequency of 5 Hz. Each acupuncture session lasted for 30 min, and patients received treatment twice daily.

Posture control training methods comprised the following: (1) improving trunk stability and head–neck position: patients were guided to adopt a seated or supine position, with therapists positioned behind them to facilitate proper head and neck posture. This involved slight flexion of the head and extension of the trunk. To stabilize the hyoid bone, therapists placed their left hand on the patient's jaw while palpating superficial large muscles such as the sternocleidomastoid and trapezius muscles, and loosening and stretching small muscles in inner and deep layers. In cases of muscle shortening, therapists addressed it by resisting jaw movements in various directions (up, down, left, and right), once daily for 15 min. This preparation was followed by head and neck control training. (2) Head and neck control training: this step encompassed passive, assistive, active, and resistive movements to achieve head-down, head-up, left–right rotation, left–right lateral flexion, and head indentation. The training involved a set of 10 movements, with 3 to 5 sets performed once daily based on the patient's adaptation. Throughout the training, patients were encouraged to avoid holding their breath and were instructed to progress at their own pace, exerting their best effort during head control exercises. (3) Shrug exercise: patients raised both shoulders as close to their ears as possible for 10 s before simultaneously lowering them to the lowest position. It was emphasized that the patient's upper limbs should remain straight during the shrugging motion. Similar to the head and neck control training, the shrug exercise consisted of a set of 10 movements, with 3 to 5 sets conducted once daily, depending on the patient's adaptability.

Water swallow test

During the treatment procedure, patients were provided with warm water at a temperature ranging from 37 to 40°C for consumption while in a seated or reclined position. Various levels were established to evaluate the patient's capacity to swallow the warm water and the incidence of coughing:

Level I: patients successfully ingested 30 mL of warm boiled water in a single attempt without issue.

Level II: patients consumed 30 mL of warm boiled water across multiple attempts without experiencing choking or coughing.

Level III: patients managed to ingest warm water in one try but encountered coughing.

Level IV: patients required more than two attempts to ingest warm water and experienced choking.

Level V: patients were unable to consume the full 30 mL of water and frequently experienced coughing.

Standardized swallowing assessment

During the initial assessment, several critical indicators were observed: (1) the patient's level of consciousness and responsiveness to external stimuli were assessed; (2) the patient's ability to control their trunk and maintain head stability was evaluated; (3) the presence of abnormal breathing patterns in the patient was noted; (4) the adequacy of the patient's lip closure was examined; (5) the symmetry of the patient's soft palate was assessed; (6) the functional status of the larynx was evaluated; (7) the integrity of the gag reflex and the patient's autonomic coughing ability were checked for normalcy.

If none of the above indicators exhibit abnormalities, the subsequent assessment is divided into two parts. In the initial stage, the patient remains seated upright and is instructed to sequentially swallow 5 mL of water, repeated three times. During this process, the swallowing function is evaluated for potential issues, such as: (1) leakage of water from the mouth; (2) ineffective movement of the larynx; (3) repetitive movements during swallowing; (4) coughing or stridor between the larynx; (5) changes in voice (e.g., weakening, hoarseness, or inability to articulate) following water consumption. Upon successful completion of the first stage, the second stage is conducted, during which the patient is required to ingest 60 mL of water at once while maintaining an upright position for further assessment of the swallowing function. This stage involves evaluating the following aspects: (1) the ability to swallow 60 mL of water and the duration taken to complete the swallowing; (2) presence of coughing during or after swallowing; (3) occurrence of laryngeal stridor during or after swallowing; (4) any changes in voice following water consumption (e.g., weakening, hoarseness, or inability to articulate); (5) presence of aspiration.

Videofluoroscopic swallowing study

To prevent alveolar deposition post-aspiration and safeguard the patient's respiratory function, it is imperative to select a water-soluble barium sulfate suspension that can be absorbed effectively. The preparation of the suspension involves mixing barium sulfate powder with an appropriate quantity of water. Typically, 200 mg of barium sulfate is combined with 286 mL of water to achieve a homogeneous solution with a concentration of approximately 60%. This suspension is subsequently blended with pastes (thickening material) to create barium with varying consistencies. Contrast foods encompass diluted liquids (resembling water), pastes (similar to yogurt), and solids (akin to pudding). For contrast imaging, the PHILIPS multifunctional digital gastrointestinal machine (Essenta Rc system) is utilized. Under this equipment, the patient sequentially swallows 2 mL, 5 mL, and 10 mL of liquid ranging from diluted to thick consistency. Subsequently, the swallowing process is meticulously observed and assessed for further evaluation.

The general scoring criteria for the degree of dysphagia are as follows:

Oral phase: (1) inability to propel food from the mouth into the throat, resulting in food spillage from the lips or requiring forceful measures to initiate swallowing, is assigned a score of 0. (2) incapacity to form cohesive food boluses for passage into the throat, leading to food dispersion in the throat, receives a score of 1. (3) if residue remains in the mouth after swallowing attempts, it is scored as 2. (4) successful transfer of food into the throat with a single swallow is assigned a score of 3. Pharyngeal phase: (1) inadequate elevation of the throat, epiglottic malfunction, incomplete closure of the soft palate arch, and insufficient swallowing reflex result in a score of 0. (2) presence of significant residual food in the pharyngeal cavity and piriform recess warrants a score of 1. (3) minimal residue storage and complete clearance achieved through repeated swallows are given a score of 2. (4) successful passage of food into the esophagus with a single swallow is assigned a score of 3.

Degree of aspiration: (1) significant aspiration without accompanying cough is scored as 0 points. (2) presence of substantial aspiration leading to choking is assigned 1 point. A small degree of aspiration without coughing receives 2 points. (3) occurrence of minor aspiration with choking is rated 3 points. (4) absence of any aspiration is scored as 4 points.

The Videofluoroscopic Swallow Study (VFSS) is assessed on a scale ranging from 0 to 10, where a score of 0 indicates severe dysphagia, 2–3 suggests moderate dysphagia, and 7–9 signifies mild dysphagia.

Statistical analysis

We established the database using Excel software, and SPSS 25.0 software was utilized for statistical analysis. Counting data were expressed as a percentage, and the measurement data was expressed as ($\bar{x}$±s). Students *T*-test, χ^2 test, Fisher's exact test, Two-way ANOVA followed Turkey's multiple comparisons test were conducted to analyze the difference. $p<0.05$ indicates that the difference is statistically significant.

Results

The demographic and clinical characteristics of the treatment group were compared with those of the control group

Table 1 presents the clinical characteristics of the participants, encompassing gender, age, mean body mass index (BMI), educational status, presence of diabetes, and stroke type. The analysis indicated no statistically significant differences both between and within groups (Table 1).

The combination of postural control and electroacupuncture significantly enhances recovery in patients suffering from dysphagia following stroke

Compared to the control group, the observation group displayed a notably higher total treatment response rate ($p<0.0001$). The total

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TABLE 1 Demographic and clinical characteristics of the study participants.

Items	Study group		p
	Control (n = 62)	Observation (n = 65)	
Gender			
Male	35 (56.45%)	40 (61.54%)	0.560
Female	27 (43.55%)	25 (38.46%)	
Age (years, mean ± SD)	65.01 ± 9.85	66.35 ± 9.84	0.640
Course (months, mean ± SD)	1.97 ± 1.60	1.73 ± 1.44	0.553
Education status			
Junior high school and below	31 (50.00%)	35 (53.85%)	0.159
Senior high school	22 (35.48%)	27 (41.54%)	
College and above	9 (14.52%)	3 (4.61%)	
Smoke			
Yes	29 (46.77%)	30 (46.15%)	0.944
No	33 (53.23%)	35 (53.85%)	
Diabetes mellitus			
Yes	14 (22.58%)	20 (30.77%)	0.298
No	48 (77.42%)	45 (69.23%)	
Stroke type			
Cerebral infarction	46 (74.19%)	51 (78.46%)	0.571
Cerebral hemorrhage	16 (25.81%)	14 (21.54%)	

Values were expressed as N (percentage [%]). $\bar{x} \pm s$ mean plus or minus the standard deviation. p values were derived from the χ^2 test, Students T-test or Fisher's exact test.

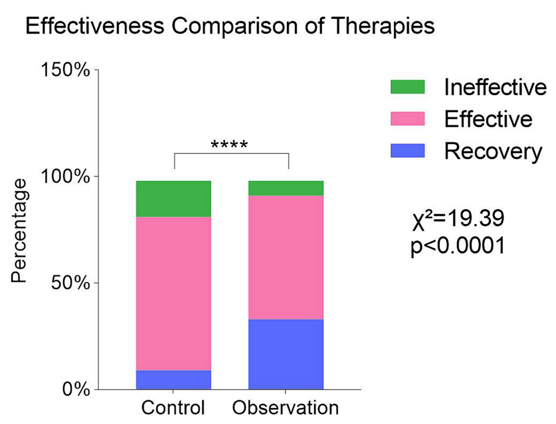

FIGURE 2
Comparison of clinical effectiveness in the treatment of dysphagia between the two groups. Values were expressed as N (percentage [%]). p value was derived from the χ^2 test. (χ^2,df) = (19.39,2), ****p < 0.0001.

effective rate was 82.26% in the control group and 92.31% in the observation group (Figure 2). These findings indicate that the observation group, receiving a combination of conventional swallowing rehabilitation training, posture control, and electroacupuncture, exhibited superior treatment efficacy compared to the control group, which received conventional swallowing rehabilitation training only.

We then assessed and compared standardized swallowing assessment (SSA), water swallow test (WST), and aspiration tests between the two groups. Both groups demonstrated some degree of recovery after treatment, with a significant difference observed between the two groups (p < 0.05, Figure 3A).

The SSA scores in both the observation group and control groups decreased after treatment, and this change was statistically significant (p < 0.0001, Figure 3A). Before treatment, there was no significant difference between the observation and control groups (p > 0.05, Figure 3A). However, after the intervention, a significant difference was observed between the control group and the observation group (Figure 3A). The WST results were compared between the two groups by calculating the number of individuals at each stage of WST in both groups. The evaluation was conducted using Two-Way ANOVA. When comparing the results before and after treatment in the two groups, a significant difference was found (p < 0.0001, Figure 3B), indicating that both treatment methods had a therapeutic effect. Furthermore, no significant difference in WST results was found before treatment between the two groups (p > 0.05, Figure 3B). However, after treatment, the observation group exhibited remarkable improvements compared to the control group (p < 0.05, Figure 3B). The combination of conventional swallowing rehabilitation training, posture control, and electroacupuncture was more effective than conventional swallowing rehabilitation training alone in improving water swallow function (Figure 3B).

Regarding the aspiration test, there was no significant difference between the two groups before treatment (Figure 4A). However, after treatment, a significant difference emerged between the two groups (p < 0.05, Figure 4B), with the observation group demonstrating greater improvement in aspiration compared to the control group. These findings indicate that patients in the observation group experienced substantial enhancements in swallowing function after treatment compared to those in the control group.

To assess the safety of the treatment and the occurrence of complications post-treatment, we evaluated fever and nutritional disorders in both groups. In contrast, among the 62 cases in the control group, 11 participants experienced fever, and 7 exhibited nutritional disorders, leading to a total incidence of 29.03%. Among the 65 cases in the observation group, only 8 participants developed fever, while none showed nutritional disorders, resulting in a total incidence of 12.13%. Statistical analysis revealed a significant difference between the two groups (p < 0.05, Figure 5). These findings suggest that the observation group had a lower incidence of fever and nutritional disorders compared to the control group, indicating the safety and benefits of the intervention in the observation group.

Discussion

Stroke is a medical condition resulting from disruptions in cerebral blood circulation, leading to ischemia and hypoxic lesion

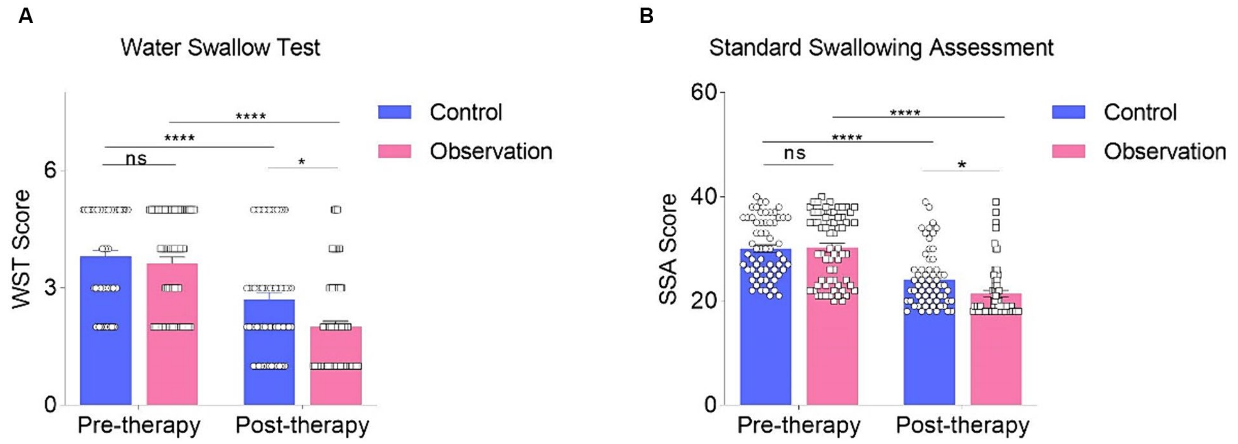

FIGURE 3
Comparison of pre- and post-treatment water swallow test **(A)** and standard swallowing assessment **(B)** was conducted using Two-way ANOVA followed by Tukey's multiple comparisons test. * $p < 0.05$, **** $p < 0.0001$, and "ns" indicates no significance.

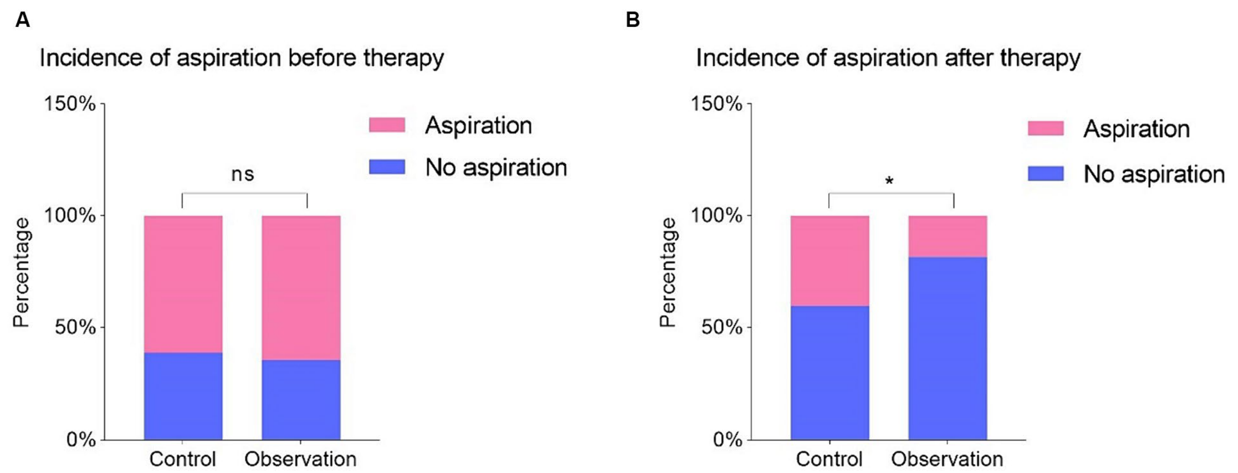

FIGURE 4
Comparison of whether there is aspiration before **(A)** and after **(B)** treatment in two groups. Values were expressed as N [percentage (%)]. *p* value was derived from the Fisher's exact test. *$p < 0.05$, and "ns" indicates no significance.

necrosis of brain tissue (14). The main pathogenesis is associated with infarction of cerebral blood vessels or sudden cerebral vascular hemorrhage, manifesting clinically as neurological loss corresponding to the affected lesion (14–17). However, when dysphagia occurs after a stroke, cerebrovascular ischemic injury damages the nerve and central functions related to swallowing (18), resulting in slow tongue movement and Swallowing muscle coordination dysfunction (19). Patients with dysphagia after stroke also suffer from additional complications, such as Speech impediment, malnutrition, Electrolyte imbalance, and aspiration (20). In particularly, aspiration can lead to lung infections, significantly impacting the prognosis of stroke patients (9, 18, 20). Therefore, effectively treating these complications after stroke is of great significance to promote physical and neurological recovery (21). However, conventional rehabilitation training and medication guidance have not shown significant efficacy in improving swallowing dysfunction (22).

Electroacupuncture, rooted in traditional Chinese medicine theory, involves stimulating meridian acupuncture points. This technique offers strong penetration and regulation of body functions (23). It enhances the immune system, balances Yin and Yang, and promotes longevity (24, 25). Previous studies indicate that acupuncture points for treating swallowing disorders after stroke include Lianquan (CV23), Tiantu (CV22), Chengjing (BL56), Chejia (ST6), Yifeng (TE17), Taixi (KI3), Fengchi (GB20), Neiguan (PC6), Sanyinjiao (SP6), Yintang (EX-HN3), Taiyang (EX-HN5), Jinjin (EX-HN12), Yuye (EX-HN13), Zusanli (ST36), Fenglong (ST40), Taichong (LR3), Xiaguan (ST7) (26). Among these, the most commonly used acupuncture points are Lianquan (CV23), Fengchi (GB20), Yifeng (TE17), Jinjin (EX-HN12), and Yuye (EX-HN13) (27).

In this study, adhering to the principle of traditional Chinese medicine theory and Meridian theory (28), the treatment for swallowing disorders after stroke primarily targets acupuncture points on the head and neck, such as Xiaguan (ST7) acupoint, and Jiache

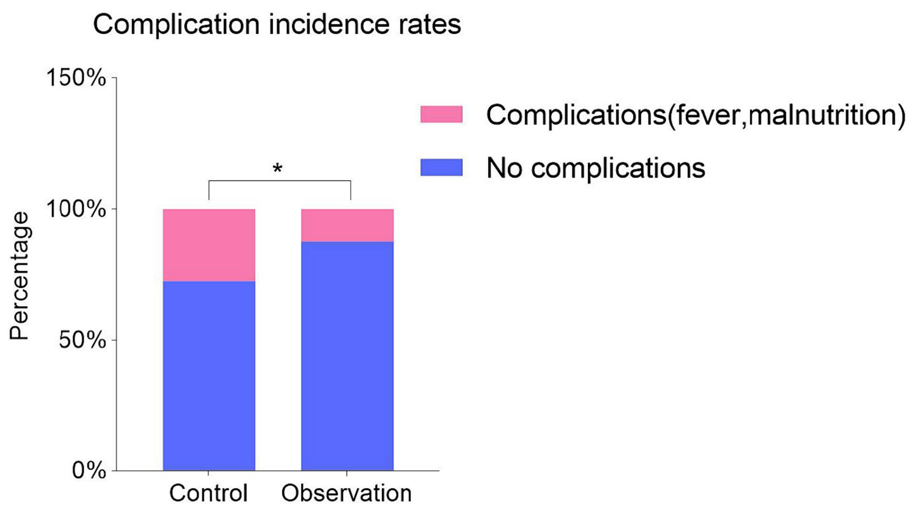

FIGURE 5
Comparison of complications (fever and malnutrition) in two groups after treatment. Values were expressed as N (percentage [%]). *p* value was derived from Fisher's exact test. * $p < 0.05$.

(ST6) acupoints, and Chengjiang (CV24) acupoint on the face (29), Lianquan (CV23) acupoint, and Jinjin (EX-HN12) acupoints, and Yuye (EX-HN13) acupoint on the tongue pharynx (30), as well as the Fengchi (GB20) and Yifeng (TE17) acupoints on the neck (31). The Chengjiang (CV24) acupoint's proximity to the mouth makes it effective in treating mouth and adjacent area disorders, which is associated with the Yin and Yang theory (32). Electroacupuncture at Lianquan (CV23) acupoints promotes the contraction of swallowing muscles and local blood circulation (33). Additionally, stimulation of the Fengchi (GB20) and the Yifeng (TE17) acupoints improves the blood rheology, peripheral microcirculation, blood flow of the brain, and skull base blood flow (33, 34), and establishes collateral circulation at the lesion to improve the blood sample supply of brain tissue as soon as possible, thereby accelerating the recovery of central nervous system function and rebuilding upper motor neurons, so that the motor nucleus of the medulla oblongata can be reasonably innervated (35). Besides, Lianquan (CV23) acupoint as a peripheral stimulation strategy, could improve the swallowing function in PSD model mice through the activation of motor cortex inputs to the NTS through the PBN. the application of EA-CV23 for the treatment of PSD by addressing how motor cortex activation benefits recovery of swallowing function via a subcortical pathway, suggesting that acupuncture treatment could be used as an effective therapeutic intervention to improve swallowing function (27).

Postural control for dysphagia after stroke is derived from the Bobath technique, which employs a neurodevelopmental approach (9). Its primary objective is to enhance nerve function by inhibiting abnormal posture by inhibiting abnormal postures, thereby promoting the development and recovery of normal posture (36). This approach proves effective in addressing stroke-related swallowing disorders as it enhances the patient's ability to control ability of body position through targeted training of the upper limbs, trunk, head, and neck (37). By holistically training multiple body parts, it creates a safe environment for patients experiencing dysphagia to eat (38), improves muscle tone, maintains the normal anatomical position of the articulation organs, and synergizes with comprehensive rehabilitation treatment to more efficiently alleviate swallowing disorders (39). The findings of this study reveal that combining posture control with electroacupuncture and conventional rehabilitation training yields notable improvements in post-stroke dysphagia, achieving an overall treatment rate of 92.31%, significantly surpassing the control group. Moreover, subsequent evaluations of indicators, including SSA, WST, and aspiration, demonstrate improved swallowing function in both groups, with the observation group displaying more significant results than the control group. Additionally, based on the evaluation of fever and nutritional disorders, the observation group exhibited superior effects.

Limitations of the study

The sample size is small and comes from a single source, consisting solely of patients from The Third Clinical Medical College of the Three Gorges University, Gezhouba Central Hospital of Sinopharm. Hence, larger-scale, multi-regional studies are warranted to validate our findings. Furthermore, no research on the efficacy of routine rehabilitation, posture control and electroacupuncture in treating dysphagia after stroke has been carried out. Therefore, the results should be interpreted with caution. Future studies should expand the sample size and groups to provide a more comprehensive understanding of the effectiveness of electroacupuncture and postural control.

Conclusion

In summary, both approaches—routine rehabilitation training and posture control combined with electroacupuncture based on routine rehabilitation training—show promising results in treating patients with swallowing disorders after stroke. These interventions enhance swallowing function and reduce the occurrence of complications such as aspiration, fever, and nutritional disorders.

Notably, the combination of electroacupuncture, posture control, and routine rehabilitation training proves to be more effective than routine rehabilitation training and provides a more favorable treatment outcome.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving humans were approved by Gezhouba central hospital of Sinopharm Ethics committee. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

YW: Writing – original draft, Validation, Supervision. ZZ: Writing – original draft, Data curation. QL: Writing – original draft, Software, Methodology. XY: Writing – original draft, Data curation. JR: Writing – original draft, Formal analysis. YC: Writing – original draft, Project administration, Investigation. HZ: Writing – review & editing, Writing – original draft, Resources, Funding acquisition, Conceptualization.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Evaluation of clinical factors predicting dysphagia in patients with traumatic and non-traumatic cervical spinal cord injury: a retrospective study

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Introduction: Dysphagia is a common complication in patients with cervical spinal cord injury (C-SCI) and can cause various pulmonary complications, such as aspiration pneumonia and mechanical airway obstruction increasing mortality and morbidity. This study evaluated the clinical factors that predict dysphagia in patients with traumatic and non-traumatic C-SCI.

Methods: Ninety-eight patients with C-SCI were retrospectively enrolled in this study and were divided into those with and without dysphagia. Clinical factors such as age, sex, tracheostomy, spinal cord independence measure, pulmonary function test (PFT) including forced vital capacity (FVC), forced expiratory volume in 1 s (FEV1) and FVC/FEV1, American Spinal Cord Injury Association score, Berg Balance Scale, and surgical approach were investigated retrospectively.

Results: Multivariate logistic regression analysis revealed that FVC and the presence of tracheostomy were significantly correlated with dysphagia in patients with C-SCI ($p < 0.05$). FVC and the presence of tracheostomy are useful tools for detecting dysphagia in patients with C-SCI.

Conclusion: Considering the results of our study, early PFTs, especially FVC, in patients with C-SCI and early initiation of dysphagia management and treatment in patients with C-SCI and tracheostomy will be advantageous in lowering the mortality and morbidity due to pulmonary aspiration in these patients.

KEYWORDS

deglutition, deglutition disorders, spinal cord injuries, vital capacity, cervical spine, dysphagia

1 Introduction

Cervical spinal cord injuries (C-SCI) are the most severe SCIs, often resulting in complete or partial loss of sensorimotor functions or even fatalities. C-SCIs are divided into traumatic and non-traumatic types (1).

Dysphagia is a swallowing function disorder characterized by the abnormal movement of liquids or a food bolus (2). In patients with C-SCI, the prevalence of dysphagia varies from 16 to 80% (3, 4). Dysphagia after a C-SCI can increase the risk of aspiration pneumonia, leading to increased mortality and morbidity (3, 4).

Complications secondary to dysphagia among patients with C-SCI include pulmonary sequelae, such as transient hypoxemia, chemical pneumonitis, mechanical airway obstruction, bronchospasm, and pneumonia (5, 6). Furthermore, dysphagia is a predisposing condition for aspiration, which may contribute to the development of pneumonia (7); moreover, complications associated with respiration are considered the leading causes of morbidity and mortality in patients with SCI (8, 9). However, despite being associated with significant morbidity, dysphagia remains under-recognized in SCI (5). Therefore, early identification of risk of dysphagia in individuals with C-SCI can prevent or reduce the development of associated adverse complications by facilitating timely diagnosis and treatment (10, 11). Furthermore, awareness of the factors associated with swallowing dysfunction among individuals with C-SCI is crucial and has important implications for medical management strategies in these patients.

Several studies have investigated risk factors for dysphagia in patients with traumatic C-SCI (10), where age, tracheostomy, injury level, severe paralysis, voice quality, peak cough flow, forced expiratory volume in 1 s (FEV1), and anterior cervical surgery were identified as risk factors for dysphagia (3, 10). However, few studies have involved patients with non-traumatic C-SCI. Therefore, this study aimed to investigate the risk factors for dysphagia in patients with both traumatic and non-traumatic C-SCI.

2 Materials and methods

2.1 Study design and population

This retrospective study was approved by the Institutional Review Board (IRB) of Ulsan University Hospital (IRB number: 22–04-037) and was performed per the Declaration of Helsinki for human experiments. The IRB of Ulsan University Hospital waived the need for informed consent due to the retrospective nature of the study. We retrospectively obtained clinical data such as sex, age, tracheostomy, spinal cord independence measure (SCIM) scores, completeness of SCI, presence of tracheostomy, neurological level, pulmonary function test (PFT) including forced vital capacity (FVC), FEV1, the ratio of FVC to FEV1 (FVC/FEV1), American Spinal Cord Injury Association (ASIA) scores, Berg Balance Scale (BBS) scores, and surgical method from patients with C-SCI at the university hospital between January 2018 and August 2022.

The inclusion criteria were as follows: (i) aged between 20 and 89 years; (ii) hospitalization for the treatment of C-SCI; (iii) weakness or sensory disturbance in both upper and lower extremities due to C-SCI; and (iv) a clear diagnosis of cervical myelopathy (or injury) using magnetic resonance imaging (MRI; C-SCI was defined as a high signal intensity at the cervical spinal cord in T2-weighted C-SCI MRI consistent with cervical myelopathy as confirmed by certified board radiologic specialists).

The exclusion criteria were as follows: (i) inability to undergo the swallowing test or PFT due to poor consciousness; (ii) dysphagia due to neurologic conditions diagnosed by neurologists, including stroke, traumatic brain injury, anoxic brain injury, brain tumor, motor neuron disease, Parkinson's disease, or Alzheimer's disease; (iii) dysphagia resulting from any other surgical or medical history, such as laryngeal or tongue cancer or vocal cord paralysis; and (iv) an interval of >28 days between the swallowing test and PFT.

2.2 Dysphagia

Patients with C-SCI were divided into dysphagia and non-dysphagia groups based on their symptoms by a rehabilitation medicine specialist (DP) with >10 years of experience in dysphagia treatment. Based on the judgment of the rehabilitation medicine specialist, a patient was assigned to the dysphagia group following positive assessments in bedside swallowing evaluations or at least one patient-reported symptom of dysphagia, such as food pressure, coughing when eating, nausea, or changes in diet. A video fluoroscopic swallowing study (VFSS) was performed on all patients with C-SCI in the dysphagia group to confirm the dysphagia. To evaluate the severity of dysphagia, penetration-aspiration scale (PAS) was used.

2.3 The spinal cord independence measure (SCIM)

The SCIM is a scale used to assess the achievement of daily function in patients with spinal cord lesions. The third version (SCIM III) contains 19 tasks organized into three subscales: self-care, respiration and sphincter management, and mobility (12). Here, we compared the effects of dysphagia in patients with C-SCI using scores of two subscales—respiration and sphincter management and mobility—and total SCIM scores. The SCIM was evaluated through observation and interviews with the patient's occupational therapists.

2.4 Pulmonary function test (PFT)

Spirometry was conducted according to American Thoracic Society guidelines (Vmax 22, SensorMedics; PFDX, MedGraphics, Dersingham, United Kingdom). The following parameters were evaluated: FVC, FEV1, and FEV1/FVC (13). All spirometric values are expressed as percentages of the predicted values (13).

2.5 American Spinal Cord Injury Association (ASIA)

The ASIA International Standards for Neurological Classification of Spinal Cord Injury form was used to evaluate spinal cord injuries (Atlanta, GA, Revised 2011, Updated 2015. Published with permission of the ASIA, Richmond, VA, United States) (14, 15). The sensory examination evaluates 28 specific dermatomes bilaterally for light touch (generally with a piece of cotton) and pinprick (generally with a clean safety pin) sensations. Each examination component was recorded for each dermatome and laterality. Grades of 0, 1, and 2 denote a lack of sensation, an impaired or altered sensation, and a normal sensation, respectively. A normal unilateral sensory examination comprises 28 dermatomes, each with 2 points for a light touch and 2 points for a pinprick, yielding a total of 112 points. A total score of 224 bilaterally was considered a fully normal sensory examination (14, 15). The motor examination graded five specific muscle groups in the upper and lower extremities, representing major cervical and lumbar myotomes. Motor strength was graded using a universal six-point scale (0–5). Motor strength was recorded bilaterally in each muscle group. The maximum bilateral motor score in a healthy individual is 100, with 50 for scoring 5/5 in all right upper and lower extremity myotomes and another 50 in the left (14, 15).

2.6 The Berg balance scale (BBS)

The BBS is a 14-item scale that quantitatively assesses balance and risk of falls in older community-dwelling adults through direct observation of their performance (16). The items were scored from 0 to 4, with a score of 0 representing an inability to complete the task and a score of 4 representing independent completion of the item (16). A physical therapist measured the BBS scores of the patients with C-SCI and calculated the global score from 56 possible points.

2.7 Surgical method

Information about the surgical approach—anterior, posterior, or combined anterior–posterior in cervical spine surgery in patients with C-SCI was noted (10).

2.8 Statistical analysis

For comparisons of the patient characteristics (age, sex, SCIM scores, ASIA scores, FVC, FEV1, FVC/FEV1 ratio, BBS, and cervical approach) between the two C-SCI groups, *p*-values were calculated using a Pearson chi-square test or Mann–Whitney U-test. Using clinical factors which showed a statistical significance in Pearson chi-square or Mann–Whitney U-tests, multivariate logistic regression analysis using the stepwise method was performed to evaluate the correlation with the presence of dysphagia. We also performed a receiver operating characteristic (ROC) analysis to investigate the sensitivity, specificity, and cutoff value of clinical factors for predicting dysphagia in patients with C-SCI. Statistical analyses were performed using MedCalc (MedCalc Software Ltd., Ostend, Belgium) and SPSS software (version 22.0; IBM Corp., Armonk, NY, United States).

3 Results

3.1 Patient characteristics

Of the 98 patients with C-SCI enrolled in this study, 72 were male and 26 were female (mean age 64.59 ± 12.53 years). The demographic data of the patients is shown in Table 1. Classification of the causes of C-SCI is presented in Table 2. Among the patients, 34 were classified into the dysphagia group and 64 into the non-dysphagia group. All patients, except for two with transverse myelitis, underwent surgery. The average interval between the VFSS and PFT in our study was 2.22 ± 1.00 days (Min–Max; 1–5 days).

TABLE 1 Characteristics of included patients with cervical spinal cord injury.

Characteristic	Dysphagia (n = 34)	Non-dysphagia (n = 64)	*p*-value
Age	66.76 ± 13.09	63.44 ± 12.28	0.382
Sex			0.504
Male, *n* (%)	26 (76.50)	46 (71.90)	
Female, *n* (%)	8 (23.50)	18 (28.10)	
SCIM			
SCIM_respiration score	21.08 ± 14.78	14.26 ± 11.15	0.152
SCIM_mobility score	8.77 ± 13.01	5.97 ± 8.70	0.407
SCIM_total score	37.38 ± 31.57	29.42 ± 21.20	0.416
ASIA			
ASIA_motor score	53.13 ± 31.28	57.63 ± 21.91	0.623
ASIA_sensory score	69.80 ± 33.01	108.43 ± 193.81	0.450
Pulmonary function test			
FVC (%predicted)	49.93 ± 27.12	66.47 ± 17.88	**0.020***
FEV1 (%predicted)	57.36 ± 29.65	72.50 ± 18.20	**0.038***
FEV1/FVC ratio	86.93 ± 8.60	82.17 ± 8.79	0.099
Cause of injury			0.187
Traumatic, *n* (%)	22 (64.70)	30 (46.90)	
Non-traumatic, *n* (%)	12 (35.30)	34 (53.100)	
Operation			0.248
Anterior approach, *n* (%)	12 (35.30)	32 (50)	
Posterior approach, *n* (%)	22 (64.70)	32 (50)	
Tracheostomy			**0.005***
Positive, *n* (%)	12 (35.30)	2 (3.1)	
Negative, *n* (%)	22 (64.70)	62 (96.9)	
ASIA impairment scale			0.099
A, *n* (%)	8 (23.5)	4 (6.30)	
B, C, and D, *n* (%)	26 (76.5)	60 (93.8)	
PAS	–	3.94 ± 2.98	

*Significant difference was noted between the two groups ($p < 0.05$). *p*-value was calculated by comparing dysphagia and non-dysphagia groups using independent *t*-test, Mann–Whitney U-test, or Pearson's chi-square test. SCIM, spinal cord independence measure; FVC, forced vital capacity; FEV1, forced expiratory volume in one second; PAS, penetration-aspiration score. Bolds indicate $p < 0.05$.

3.2 The difference in parameters between the dysphagia and non-dysphagia groups

In the independent Pearson chi-square test or Mann–Whitney U-test, FVC, FEV1, and the presence of tracheostomy were significantly different between the dysphagia and non-dysphagia groups ($p < 0.05$; Table 1). There was no significant between-group difference in the age, sex, SCIM score, cause of injury, ASIA motor and sensory score levels, and completeness of SCI ($p > 0.05$; Table 1).

In the multivariate logistic regression analysis, FVC and the presence of tracheostomy were found to be significantly associated with dysphagia in patients with C-SCI ($p < 0.05$; Table 3).

In the ROC curve analysis, the area under the ROC curve of FVC for the presence of dysphagia was 0.714 (95% confidence interval 0.558–0.840; $p = 0.0386$). The optimal threshold for detecting dysphagia in patients with C-SCI was determined to be a predicted maximal Youden index of ≤48% (sensitivity, 64.29%; specificity, 86.67%; see Figure 1).

4 Discussion

Several studies investigating the risk factors for dysphagia in patients with traumatic C-SCI have identified age, tracheostomy, severe paralysis, voice quality, PFT results (peak cough flow or FEV1), and anterior cervical surgery as risk factors for dysphagia in patients with traumatic C-SCI (3, 10). To the best of our knowledge, no previous study has investigated the risk factors for dysphagia involving both patients with traumatic and non-traumatic C-SCI. The results of this study suggest that the presence of tracheostomy and FVC ≤48% of predicted are significantly associated with dysphagia (sensitivity, 64.29%; specificity, 86.67%). The reason our study had fewer risk factors for dysphagia compared with previous studies is thought to be the inclusion of both traumatic and non-traumatic C-SCI. The FVC is correlated with the inspiratory muscles' power (17). Although the inspiratory muscles comprise several muscle groups; the main inspiratory muscles are the diaphragm, which is innervated by the phrenic nerve originating from the C3-C5 root (18). However, there were no statistically significant between-group differences in the SCIM motor and sensory scores, completeness of SCI, or the ASIA motor and sensory scores, indicating the severity of SCI. Considering this, these same spinal pathways (e.g., for intercostals and accessories) are also important in swallowing, and additionally affect swallow-breathing coordination (19). In addition, the relationship between swallowing and breathing may have influenced these results, as precise coordination between the two is important to prevent pulmonary aspiration (20). Although the exact neural processes by which these behaviors (breathing and swallowing) are coordinated are not well understood, it is hypothesized that swallowing and breathing are coordinated to occur at specific times relative to one another to minimize the risk of aspiration by a common control system present in the brainstem (21). In addition, FEV1, which measures the instantaneous expiratory volume for 1 s and was reported to be correlated with dysphagia in traumatic C-SCI, may be affected more by other clinical factors such as airway obstruction than FVC due to its shorter measurement time. Therefore, PFT parameters, especially FVC, which can represent breathing ability and ventilation to some extent, may be associated with the presence of dysphagia in patients with C-SCI.

In addition, we found that the presence of tracheostomy was significantly different between dysphagia and non-dysphagia groups in patients with C-SCI. In many previous studies, tracheostomy has been associated with swallowing difficulty, such as increased incidence of aspiration, and several possible mechanisms have been proposed. These are, first, the reduction of laryngeal elevation due to the tethering of the larynx by the tracheostomy tube (22); second, occlusion of the pharyngeal pathway by the tracheal tube cuff (23); third, loss of protective reflexes and desensitization of the larynx due to chronic air diversion (24); fourth, uncoordinated laryngeal closure due to chronic upper airway bypass (25). Considering this, early management and treatment of dysphagia in patients with C-SCI and tracheostomy will help lower the mortality and morbidity due to pulmonary aspiration.

In our previous study (26), we examined the correlation between various clinical data (age, sex, severity of SCI, PFT results, BBS, and surgical methods) and the severity of dysphagia in 56 patients with traumatic C-SCI distinct from those included in the current study. We found that the anterior surgical approach was the only significant factor associated with dysphagia severity in these patients. In the previous study (26), we recommended that clinicians should pay particular attention to the potential for occurrence of dysphagia in patients who undergo anterior cervical surgery. Unlike our previous study (26), the present study included both traumatic and non-traumatic patients with C-SCI. Furthermore, instead of assessing dysphagia severity, we evaluated the presence or absence of dysphagia. We compared patient characteristics between the traumatic and non-traumatic C-SCI groups. The current study revealed that FVC results and the presence of tracheostomy are associated with the development of dysphagia in patients with C-SCI.

In this study, the interval between PFT and VFSS was set relatively short (1–5 days). Recovery in patients with C-SCI varies individually, and the speed of recovery can be influenced by various factors. Therefore, assessing lung function and swallowing ability over time is

TABLE 2 Classification of causes of cervical spinal cord injury in patients.

Cause	*n* = 98
Traumatic	70 (71.4%)
Non-traumatic	
Transverse myelitis	4 (4.1%)
Ossification of posterior longitudinal ligament	20 (20.5%)
	98 (100%)

TABLE 3 Multivariate logistic regression analysis (with the Stepwise method) of the presence of dysphagia and FVC and tracheostomy in the patients with cervical spinal cord injury.

	B	Standard error	*p*-value	Exp(B)	95% CI	
					Lower bound	Upper bound
FVC	−0.007	0.003	**0.017***	−0.326	−0.012	−0.001
Tracheostomy	0.572	0.178	**0.003***	0.422	0.213	0.931

CI, confidence interval; FVC, forced vital capacity. * < 0.05. Bolds indicate $p < 0.05$.

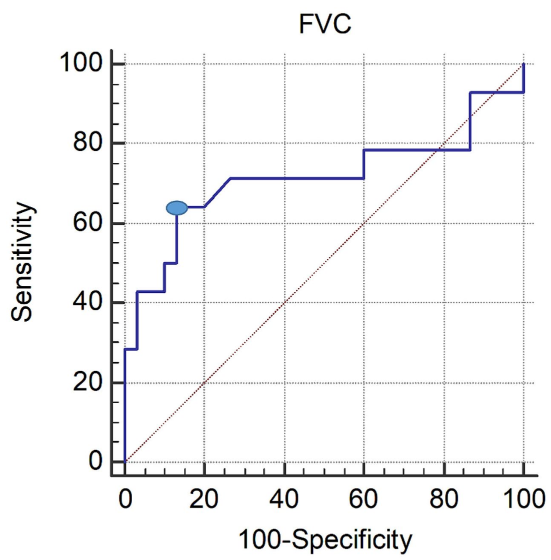

FIGURE 1
ROC curve of the FVC in patients with dysphagia. The optimal cutoff value (dot on the curve) of the FVC, obtained from the maximal Youden index, was ≤48% predicted (AUC, 0.737; 95% CI, 0.530–0.943; $p = 0.024$; sensitivity 69.23%; specificity 84.21%). ROC, receiver operating characteristic; FVC, forced vital capacity; AUC, area under the ROC curve; CI, confidence interval.

crucial in determining accurate diagnosis and treatment direction. Performing these tests at the appropriate time allows for the evaluation of respiratory and nutritional status and the initiation of appropriate treatment. This contributes to the improvement in overall recovery and quality of life for patients.

Our study has a few limitations. First, this was a retrospective review of medical records, which had the inherent limitations of small sample sizes and heterogeneity. Comparison with previous prospective studies (27, 28) suggests that selective bias may not be completely excluded. Second, the number of patients with non-traumatic C-SCI was relatively small, which may be due to differences in prevalence, and further studies with a larger number of patients with non-traumatic C-SCI are necessary. Third, in our study, VFSS was not performed on all patients with C-SCI. Screening patients with dysphagia using the bedside swallowing test and clinical symptoms may be problematic because patients with asymptomatic dysphagia may be missed. However, to avoid unnecessary long-term radiation exposure due to continuous fluoroscopy during VFSS (10 ~ 20 min), in our hospital, we tried to minimize the above limitation by observing the clinical symptoms related to dysphagia daily and performing the bedside swallowing test at the following two time points: at the time of the first consultation with the department of rehabilitation medicine and immediately upon transfer to the department of rehabilitation medicine. At these two points, based on the judgment of the rehabilitation medicine specialist with more than 10 years of experience in the evaluation and management of dysphagia, patients with C-SCI with positive assessments in bedside swallowing evaluations or reports of at least one symptom of dysphagia, such as food pressure, coughing when eating, nausea, or changes in the diet at any time, underwent VFSS. Patients showing penetration or aspiration in VFSS (PAS ≥2) were assigned to the dysphagia group, and the management and treatment were prescribed. To evaluate the clinical factors correlated with dysphagia in traumatic and non-traumatic C-SCI more precisely, however, further prospective studies with routine VFSS in C-SCI with numerous patients with C-SCI may be necessary. Fourth, this study was conducted at a single tertiary hospital, and future multicenter studies could address the potential bias in the findings reported in this study. Therefore, further prospective studies with larger sample sizes are needed for more accurate generalization.

In conclusion, FVC and the presence of tracheostomy are useful tools for detecting dysphagia in patients with C-SCI. Considering the results of our study, early PFTs, especially FVC, in patients with C-SCI and early initiation of dysphagia management and treatment in patients with C-SCI and tracheostomy will greatly benefit in lowering the mortality and morbidity due to pulmonary aspiration.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by the Institutional Review Board (IRB) of Ulsan University Hospital. The studies were conducted in accordance with the local legislation and institutional requirements. The ethics committee/institutional review board waived the requirement of written informed consent for participation from the participants or the participants' legal guardians/next of kin because retrospective nature of the study.

Author contributions

J-WC: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Resources, Validation, Visualization, Writing – original draft, Writing – review & editing. DK: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Resources, Validation, Visualization, Writing – original draft, Writing – review & editing. SJ: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Resources, Validation, Visualization, Writing – original draft, Writing – review & editing. DP: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Resources, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. MC: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Resources, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Exploring the rules of related parameters in acupuncture for post-stroke dysphagia based on data mining

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Background: Post-stroke dysphagia (PSD) affects the efficacy and safety of swallowing, causing serious complications. Acupuncture is a promising and cost-effective treatment for PSD; however, as the number of randomized controlled trials increases, scientific analysis of the parameters and acupoint prescription is required. Therefore, this study aimed to explore the effects of acupuncture on parameters related to post-stroke dysphagia (PSD).

Methods: We searched the Cochrane Library, PubMed, Embase, Web of Science, China National Knowledge Infrastructure, Wanfang Database, Chinese Biomedical Literature, and Chongqing VIP Database for randomized controlled trials of acupuncture for PSD in the last 15 years and relevant parameters were analyzed using data mining techniques.

Results: In total, 3,205 records were identified, of which 3,507 patients with PSD were included in 39 studies. The comprehensive analysis demonstrated that the closest parameter combinations of acupuncture on PSD were 0.25 mm × 40 mm needle size, 30 min retention time, five treatments per week, and a 4-week total course of treatment. Additionally, the gallbladder and nontraditional meridians, crossing points, and head and neck sites are the most commonly used acupoint parameters. The core acupoints identified were GB20, RN23, EX-HN14, Gongxue, MS6, SJ17, EX-HN12, EX-HN13, and the commonly used combination of EX-HN12, EX-HN13, GB20, and RN23.

Conclusion: This study analyzed the patterns of PSD-related needling and acupoint parameters to provide evidence-based guidelines for clinical acupuncturists in treating PSD, potentially benefitting affected patients.

KEYWORDS

acupuncture, stroke, dysphagia, parameter, data mining RN23, EX-HN14, Gongxue, MS6

1 Introduction

Dysphagia is a common complication of stroke, and the incidence of post-stroke dysphagia (PSD) ranges from 37–78% (1). PSD impairs the efficacy and safety of swallowing, causing a significant psychological burden and serious complications, such as dehydration, malnutrition, aspiration pneumonia, readmission, and even death (2). In addition to complications and reduced quality of life, PSD increases healthcare costs. A systematic review reported the costs of $16,900 for PSD and $27,600 for pneumonia (3). A prospective cohort study demonstrated that 32.3% of patients with ischemic stroke experienced dysphagia after 7 days, and 80.5% had dysphagia at hospital discharge (4). Thus, managing, preventing, and treating PSD to reduce its incidence, complications, and healthcare costs are current challenges.

Currently, clinical practice guidelines recommend dietary, behavioral (acupuncture), nutritional, pharmacological, and neurostimulatory interventions for PSD (5). Pharmacological treatments can only provide moderate symptom control and often lead to adverse reactions. Although nutritional supplements and neuro-dietary modulation hold clinical significance, the absence of effective markers for malnutrition and challenges in assessing nutritional status are major issues (6). Acupuncture is a promising and cost-effective treatment. A high-quality RCT demonstrated that the cost per quality adjusted life year gained from acupuncture was only 4,241 pounds, well below the commonly accepted threshold of 20,000 pounds. Moreover, sensitivity analyses confirmed a greater than 90% probability of cost-effectiveness, highlighting its economic and therapeutic value (7). Previous meta-analyses have shown that acupuncture re-establishes swallowing function and effectively improves quality of life in patients with PSD (8, 9). Compared to non-acupuncture treatments (swallowing training, medication), acupuncture treatment has a higher efficacy rate (RR, 1.33; 95% confidence interval, 1.25 to 1.43) (8). However, no studies have explored the parameters related to acupuncture in PSD, including the details of acupuncture, core acupoints, and potentially effective combinations of acupoints. Additionally, the standards for reporting interventions in clinical trials of acupuncture (STRICTA) guideline recommend the detailed reporting of the acupuncture parameters implementation process to improve the evaluation of acupuncture efficacy and promote the replication and dissemination of acupuncture studies (10). Therefore, as the number of acupuncture-related randomized controlled trials (RCTs) expands, scientific analyses of the parameters and acupoint prescriptions related to acupuncture for PSD are required.

This study aimed to comprehensively assess the clinical parameters of acupuncture on PSD by conducting a secondary analysis of the literature. Data mining techniques (descriptive analysis, association rules, and cluster analysis) were used to explore the effects of acupuncture on PSD-related parameters, develop standard and effective prescriptions for PSD, reduce confounding factors in the study, and improve the efficacy of acupuncture treatment and the quality of clinical studies.

2 Methods

2.1 Criteria for considering reviews for inclusion

This systematic review aimed to explore the effects of acupuncture on parameters related to PSD. RCTs on acupuncture interventions for patients with acute or recovering PSD without language restrictions were included. To maintain the rigor of this study, pseudo-randomization, quasi-randomization, experimental studies, reviews, case reports, letters, studies with incomplete data, and publications with duplicate data (data extracted from the most recent literature) were excluded. Studies that did not report any of the parameters of interest were excluded.

All qualifying studies included a control group as a minimum requirement. Furthermore, patients with PSD who underwent acupuncture in the included studies had positive outcomes compared to controls.

2.2 Types of participants

PSD is the most frequent condition in which stroke damages the swallowing network, leading to dysphagia (5). Patients with acute (within 7 days) and recovery (within 6 months) PSD were included. Patients with functional dysphagia and dysphagia due to esophageal cancer, laryngeal cancer, or inflammation were excluded. No restrictions based on the type of stroke (hemorrhage or ischemia), location of the lesion (brainstem or non-brainstem), side of the lesion (unilateral or bilateral), age, sex, or geographic region imposed.

2.3 Types of interventions and comparisons

Acupuncture therapies in the intervention group included manual acupuncture, electroacupuncture, and conventional physical therapy techniques, whereas those in the control group included rehabilitation, sham acupuncture, placebo acupuncture, or drugs. Studies using moxibustion, acupoint burrowing, auricular acupuncture, and non-invasive interventions (Chinese herbs and tuina) were excluded. No clinical trials comparing different acupuncture methods or acupoint-taking protocols were considered.

2.4 Types of outcome measures

Study inclusion in the analysis depended on providing at least one of the following standardized and validated dysphagia scales: the Penetration Aspiration Scale, Functional Oral Intake Scale, Water Swallow Test, Standardized Swallowing Assessment, Videofluoroscopic Swallowing Study, Modified Mann Assessment of Swallowing Ability, Dysphagia Outcome Severity Scale and Swallowing-Quality of Life. Studies that reported only physical or chemical examinations were excluded. The analysis time point was set at the end of all scheduled treatment sessions.

2.5 Information sources and search

All articles describing acupuncture and PSD were obtained from the Cochrane Library, PubMed, Embase, Web of Science, China National Knowledge Infrastructure, Wanfang Database, Chinese Biomedical Literature, and Chongqing VIP Database between December 2009 and December 2023 in English and Chinese.

The PubMed and Web of Science search strategies according to population, intervention, control, and outcomes (PICO) format are

Abbreviations: PSD, Post-stroke dysphagia; STRICTA, Standards for reporting interventions in clinical trials of acupuncture; RCTs, Randomized controlled trials; PICO, Population, intervention, control, and outcomes; SSA, Standardized swallowing assessment; WST, Water swallow test; SWAL-QOL, Studies reported swallowing-related quality of life; MCC, Maximal clique centrality; MNC, Maximum neighborhood component; MCODE, Molecular complex detection tool.

presented in Table 1. For the other electronic databases, the search approach was adjusted as required.

2.6 Data extraction and selection

After eliminating duplicates, two independent reviewers (MW and WS) screened the literature against the inclusion eligibility criteria (checking the title, abstract, and full text of the papers to identify eligible trials) and cross-checked the screening results. In cases of disagreement, a third reviewer (XW) made the final decision on whether the study should be included.

Data for further analysis were extracted from the included studies: demographic information (title, authors, and year), sample characteristics (age, sex, and patient volume), interventions, and acupuncture parameters (needle type, frequency of treatment, duration of session, duration of stimulation, and acupuncture points).

To evaluate the data on an intention-to-treat basis, missing data were contacted through the corresponding author's e-mail addresses. If missing data could not be obtained, the currently available literature was excluded.

2.7 Literature quality assessment

Two independent reviewers (MW and WS) autonomously evaluated potential biases using "yes," "no," or "unclear," including selection, performance, detection, attrition, reporting, and other biases.

2.8 Data synthesis and analysis

2.8.1 Descriptive statistics

Microsoft Excel 2023 was used to conduct statistical descriptive analyses of the relevant parameters in the included studies, including the type of needling, duration of single stimulation, weekly treatment frequency, total treatment duration, and acupoint frequency analysis (acupoint usage, meridian usage, specific acupoint usage, and acupoint location distribution).

2.8.2 Association rule analysis

In this study, the 15 most frequently used acupoints were analyzed. Apriori association rule analysis was conducted using the "arules" and "arulesViz" packages in R software (version 4.3.0). The strength of the association rules met the following criteria: minimum support threshold of 20% and minimum confidence threshold of 90%. Furthermore, if the uplift factor was greater than one, the effect was considered stable.

2.8.3 Complex network analysis

The acupuncture prescriptions were transformed into a vector format, and the vectors were entered into a prescription table in a 0/1 format. These standardized vectors were then imported into SPSS Modeler 18.0, resulting in an association rule stream that included acupoint nodes and their weights. Subsequently, this association rule stream was loaded into Cytoscape 3.10.1 to generate a visual association network of effective acupoints for PSD treatment. Thicker connection lines represent more frequently used acupoints.

TABLE 1 Search strategy according to a focused question (PICOS).

Database	Search equation
PubMed	("Acupuncture" [Mesh] OR "Electroacupuncture" OR "Manual acupuncture" OR "Hand acupuncture") AND ("Stroke" [Mesh] OR "Cerebral ischemia" OR "Cerebrovascular disease") AND ("Deglutition Disorders" [Mesh] OR "Dysphagia" OR "Swallowing Disorder") AND ("Randomized controlled trial" OR "Randomized")
Web of Science	#1 TS = (Acupuncture) OR AB = (Electroacupuncture) OR AB = (Manual acupuncture) OR AB = (Hand acupuncture) #2 TS = (Stroke) OR AB = (Cerebral ischemia) OR AB = (Cerebrovascular disease) #3 TS = (Deglutition Disorders) OR AB = (Dysphagia) OR AB = (Swallowing Disorder) #4 TS = (Randomized controlled trial) OR AB = (Randomized) #5 #1 AND #2 AND #3 AND #4

2.8.4 Clustering and correlation analysis

R software (version 4.3.0) was used for the clustering and correlation analyses of the 20 acupoints with the highest frequency. Clustering and correlation heatmap analyses were based on the Phi correlation coefficient, which measures the strength and direction of the relationships between binary variables.

3 Results

3.1 Study selection and characteristics

In total, 3,205 relevant studies were identified. Of these, 925 were duplicates, and 1729 were excluded after screening the titles and abstracts. A total of 512 articles were excluded after full-text evaluation, and 39 articles (11–49) were included for further analysis. The study screening process is visualized using a flowchart (Figure 1).

Detailed characteristics of the included studies are presented in Table 2. Only 3 of the 39 studies were published in English (13, 31, 41), while the rest were published in Chinese. All of these studies were conducted in China over the last 15 years. A total of 3,507 patients with PSD were included, with sample sizes ranging from 21–125 patients. In the treatment group, 27 studies (12–21, 23–25, 27, 28, 30, 31, 33, 35–37, 39, 40, 42, 44, 45, 49) used acupuncture combined with rehabilitation, and seven studies used acupuncture alone (26, 34, 38, 41, 46–48). For the control group, 32 studies used rehabilitation (12–21, 23–25, 27, 28, 30, 31, 33–37, 39–42, 44–49), whereas the remaining studies used sham acupuncture, medication, repetitive transcranial magnetic stimulation, and general nursing care. Regarding numerous outcome indicators, 13 and 27 studies reported the standardized swallowing assessment (SSA) (15–17, 20, 22, 23, 26–28, 31, 40, 46, 47) and water swallow test (WST) (11, 12, 16, 18–21, 23–25, 28–39, 41–43, 45, 47, 49) as valid outcomes, respectively. Nine studies (15, 19, 20, 24, 31, 36, 40, 42, 46) reported swallowing-related quality of life (SWAL-QOL) as an outcome indicator in patients with PSD.

All 39 included RCTs reported positive outcomes, supporting the therapeutic effectiveness of acupuncture for patients with PSD.

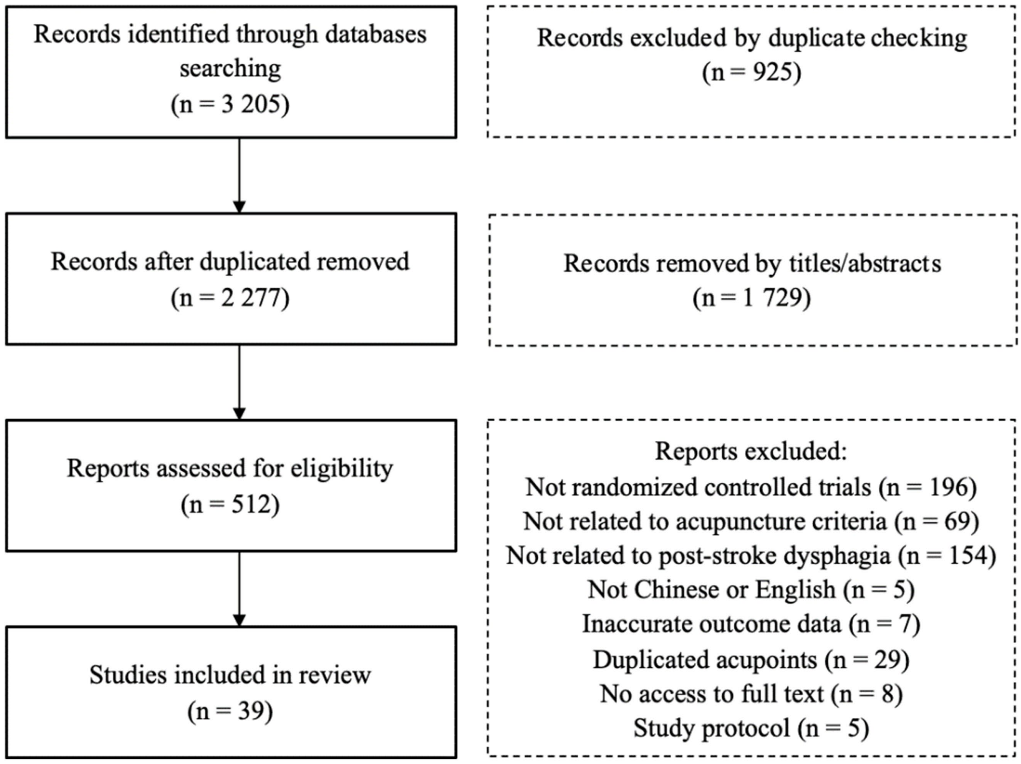

FIGURE 1
Flowchart of the study identification and selection process.

3.2 Risk of bias assessment

All studies reported randomized allocation methods and no pseudo- or quasi-RCTs. Eight studies reported allocation concealment (13, 15, 31, 35, 39–42), except for one, which had a high risk of bias (46), whereas the remaining studies did not report allocation concealment, giving an unclear risk of bias. The acupuncturists were almost impossible to blind, and the remaining literature had a high risk of bias, except for two English-language papers, which were unclear about the risk of bias (13, 31). Seven studies reported a detection bias (13, 14, 31, 40–42, 46), and the remaining studies reported an unclear risk of bias. In the incomplete outcome and selective reporting section, the remaining studies gave a low risk of bias, except for one that gave unclear selective reporting (41). Regarding other biases, 12 studies reported an unclear risk of bias (11, 12, 24, 27, 29, 30, 35, 38, 43–45, 47), while the remaining studies reported a low risk of bias (Figure 2).

3.3 Frequency of needle type

Eleven needle types were analyzed in 39 studies with a frequency of 27 uses. However, 21 studies did not report the needle type, which may have affected the RCT replication. Notably, one study reported needle-type unavailability, which was 0.20–0.25 mm × 25–75 mm. The most used needle type in these studies was 0.25 mm × 40 mm with a frequency of eight uses, accounting for 30% of the total frequency. Six needle types were used at a frequency of 1 and 6 for almost more than half of the needle types (Table 3).

3.4 Retention time

Five retention times were observed in 39 studies, with the longest being 30 min and the shortest being 5 min. The 30 min retention time was used as frequently as 24 times or 71%. In clinical practice, the choice of 30 min of acupuncture intervention for PSD may have the intended effect. Although only 5 min occurred once, the study yielded positive results. Notably, retention time was not available in six studies (Table 4).

3.5 Frequency of treatment

The results of the frequency analysis of treatments in the 39 studies showed four types, which were 5, 6, 7, and 3 days per week. The highest frequency was 5 days per week, and the lowest frequency was 3 days per week. Except for 3 days per week, the frequency share of the other treatments was approximately 0.3. However, the frequency of treatment was not available in 3 out of 39 studies (Table 5).

3.6 Total treatment duration

A total of 39 studies included five acupuncture interventions for PSD, all of which reported the total treatment time. Four weeks of intervention were used as frequently as 22 times, which was more than half of all frequencies. Six and 8 weeks total intervention time frequency of 6 times is nearly 20%. This may indicate that the PSD

TABLE 2 Characteristics included trials investigating acupuncture in patients with post-stroke dysphagia.

Author (year)	Age, mean, y (I/C)	Sample size (I/C)	Intervention group	Control group	Frequency of retention time (min)	Frequency of treatment (per week)	Frequency of total treatment duration (week)	Outcomes of interest
Chen et al. (13)	62.52/64.06	125/125	MA + Re	Re	30	6 days	3	VFSS, FMA, NIHSS
Xia et al. (40)	65.3/66.1	62/62	MA + Re	Re	30	6 days	4	SSA, DOSS(VFSS), MBI, SWAL-QOL
Liu et al. (31)	67.0/67.1	48/49	MA + Re	Re	30	5 days	8	WST, SSA, SWAL-QOL, RSST
Ren et al. (33)	63.72/64.10	45/45	MA + rTMS	rTMS	20	6 days	4	FOIS, NIHSS, FMA, Barthel Index
Zhang et al. (46)	61.1/60.7	36/36	MA	Re	30	6 days	3	SSA, SWAL-QOL, VFSS
Yu et al. (42)	71/71	21/21	MA + Re + NMES	Re + NMES	30	5 days	3	VDS, WST, FOIS, SWAL-QOL
Huang et al. (23)	NA	50/50	MA + Re	Re	NA	5 days	3	WST, SSA
Huang and Song (22)	57/57	49/49	MA + Me	Me	40	6 days	4	SSA, FDA
Huang and Song (22)	58/57	49/49	MA + Me	Me	40	6 days	4	SSA, FDA
Wang et al. (36)	59.62/60.33	40/40	EA + Re	Re	30	5 days	4	WST, MMASA, SWAL-QOL, sEMG
Chen et al. (15)	62.0/63.0	50/47	MA + Re	Re	30	5 days	4	SSA, SWAL-QOL
Li et al. (28)	61.9/63.6	40/40	MA + Re	Re	30	6 days	4	PAS(FEES), WST, SSA
Wang et al. (37)	65.32/64.76	30/30	MA + Re	Re	30	5 days	4	WST
Fang (18)	58/58	30/30	MA + Re	Re	30	6 days	4	WST
Tong et al. (35)	63/62	30/30	MA + Re	Re	30	5 days	6	WST
Yu et al. (43)	54.30/55.50	30/30	MA + GN	GN	20	5 days	4	WST
Wu et al. (39)	66/68	75/73	MA + Re	Re	30	5 days	4	WST
Li et al. (25)	NA	30/30	MA + Re	Re	30	NA	4	WST
Liu et al. (30)	58.65/58.65	100/100	MA + Re	Re	10	NA	6	WST
Chen and Hu (16)	61.80/ 60.09	35/35	MA + Re	Re	30	5 days	8	WST, SSA, FOIS
Dai et al. (17)	60.47/ 60.74	60/60	MA + Re	Re	10	7 days	8	SSA, NIHSS
Gou and Wang (19)	58.96/ 59.15	44/44	MA + Re	Re	20	7 days	4	WST, FDS, SWAL-QOL
Yuan et al. (44)	62.2/59.9	11/14	MA + Re	Re	30	5 days	2	SSS, MMASA
Zheng and Sun (47)	62.57/61.26	45/45	MA	Re	NA	6 days	4	WST, VFSS, SSA
Zhu et al. (49)	63/64	40/40	MA + Re	Re	30	5 days	4	WST, VFSS
Su and Li (34)	63.77/68.80	30/30	MA	Re	30	6 days	4	WST, FDS, MBI
Bai (12)	63.34/63.15	40/40	MA + Re	Re	20	7 days	4	WST
Li and Yu (27)	58/59	30/30	MA + Re	Re	30	6 days	4	SSA
Zhou et al. (48)	65/64	30/30	MA	Re	30	5 days	2	VFSS
Yu and Hu (45)	63/64	40/38	MA + Re	Re	20	7 days	3	WST
Li et al. (29)	59/62	30/30	MA + GN	GN	NA	7 days	2	WST
Pei (32)	62.12/61.94	39/39	MA + GN	GN	NA	7 days	2	WST
Li et al. (26)	66.4/ 66.6	30/30	MA + Re	Sham MA + Re	30	5 days	4	PAS, SSA, VFSS,
Ji et al. (24)	64.25/63.71	32/32	MA + Re	Re	NA	7 days	2	WST, FDS, SWAL-QOL
He (21)	58.6/ 59.3	33/33	MA + Re	Re	30	7 days	4	WST
Han and Gao (20)	62/63	30/30	MA + Re	Re	30	7 days	2	SSA, WST, SWAL-QOL
Chen and Ni (14)	76.58/ 73.90	40/40	MA + Re	Re	30	7 days	3	RBHOMS, FPBVS
Bahetibek et al. (11)	63.96 /63.87	65/65	MA + GN	GN	5	NA	8	WST

(Continued)

TABLE 2 (Continued)

Author (year)	Age, mean, y (I/C)	Sample size (I/C)	Intervention group	Control group	Frequency of retention time (min)	Frequency of treatment (per week)	Frequency of total treatment duration (week)	Outcomes of interest
Wei et al. (38)	58.93 /58.25	39/40	MA	Sham MA	NA	6 days	4	WST
Xie et al. (41)	66.0/68.4	70/70	MA	Re	30	5 days	4	WST

MA, Manual Acupuncture; Re, Rehabilitation; rTMS, repetitive Transcranial Magnetic Stimulation; Me, Medicine; EA, Electropuncture; GN, General Nursing; WST, Water Swallow Test; SSA, Standardized Swallowing Assessment; SWAL-QOL, Swallowing-Related Quality of Life; VFSS, Video-fluoroscopic Swallowing Study; DOSS, Dysphagia Outcome Severity Scale; MBI, Modified Barthel Index; FOIS, Functional Oral Intake Scale; FEES, Fiberoptic Endoscopic Evaluation of Swallowing; PAS, Penetration Aspiration Scale; SSS, Swallow Severity Scale; FDS, Fujishima Dysphagia Scale; RBHOMS, Royal Brisbane Hospital Outcome Measure for Swallowing; FPBVS, Food Properties and Beverage Viscosity Scores; FMA, Fugl-Meyer Assessment; NIHSS, National Institutes of Health Stroke Scale; FDA, Frenchay Dysarthria Assessment-2; RSST, Repetitive Saliva-Swallowing Test; sEMG, Surface Electromyography; MMASA, Modified Mann Assessment of Swallowing Ability; NMES, Neuromuscular Electrical Stimulation; VDS, Videofluoroscopic Dysphagia Scale; NA, Not Available.

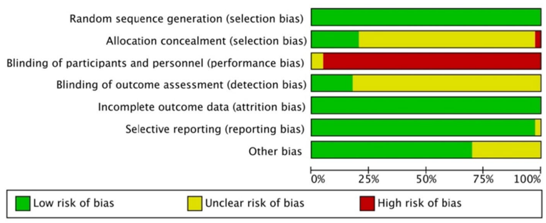

FIGURE 2
Risk of bias assessment.

recovered better and for a shorter duration with acupuncture interventions (Table 6).

3.7 Acupoint distribution

Forty PSD-eligible intervention prescriptions, 71 acupoints, and 322 occurrences were identified in 39 studies. The top 15 acupoints included Fengchi (GB20), Lianquan (RN23), Jinjin (EX-HN12), Yuye (EX-HN13), Tunyan (new acupoint), Yifeng (SJ17), Yiming (EX-HN14), Wangu (GB12), Gongxue (new acupoint), Zhiqiang (new acupoint), Fengfu (DU16), Anterior oblique line of vertex-tempora (MS6), Baihui (DU20), Wai jinjin (Wai EX-HN12), and Wai yuye (Wai EX-HN13) (Table 7 and Figure 3A).

Of the 40 acupoint prescriptions, 46 were used by 14 conventional meridians. Specifically, there were 12 conventional meridians, 1 Conception Vessel (RN), 1 Governor Vessel (DU), and nonconventional meridians. Notably, nonconventional meridian acupoints accounted for the highest number of acupoints (19) and were used as frequently as 80 times. Although not conventional, the high frequency of new acupoints indicates that clinical experience should not be ignored. In the meridian analysis, LU, LR, and HT meridians were all used twice, and all were one acupoint. Although GB had a frequency of 45 usages, only two acupoints (GB20 and GB12) may be potentially valuable for acupuncture interventions in PSD. Moreover, the SJ, ST, LI, BL, PC, SP, KI, LU, LR, and HT meridians were used <5% of the total frequency, and these meridians may be less appropriate for acupuncture intervention in PSD (Table 8).

Of the 71 acupoints, 44 were specific, and 10 contained more than one attribute. Specifically, PC6, LU7, SJ5, and SP4 were Luo-connecting points and eight confluent points. RN12 possesses the attributes of eight influential points: the front Mu and crossing points. The frequency, percentage, and highest number of acupoints used as crossing points in the specific-point analysis were 123, 38%, and 15, respectively. Acupuncturists should combine the crossing points in clinical practice to achieve better therapeutic outcomes. The Yuan primary point and Luo connecting point also had a moderate frequency in this study: 13 and 9 times, respectively. Notably, the frequencies of the eight influential points, lower He-Sea points, front Mu points, and back Shu points were <0.01, indicating that clinical acupuncturists seldom considered these specific acupoints for therapeutic PSD (Table 9).

In the acupoints distribution analysis, the head, face, and neck were the most selected areas, with frequencies, percentages, and acupoints of 279, 87%, and 47, respectively. This area is consistent with the clinical symptoms of patients with PSD, indicating that acupuncturists tend to consistently select the acupoint locations. The chest, abdomen, and back had very few acupoints, nine in total, and these locations were not considered, possibly because of disease specificity (Table 10).

TABLE 3 Frequency of needle type.

No.	Needle type	Frequency	Percentage
1	0.25 mm × 40 mm	8	0.30
2	0.30 mm × 40 mm	5	0.19
3	0.25 mm × 25 mm	4	0.15
4	0.25 mm × 50 mm	2	0.07
5	0.35 mm × 75 mm	2	0.07
6	0.20 mm × 25 mm	1	0.04
7	0.30 mm × 50 mm	1	0.04
8	0.30 mm × 70 mm	1	0.04
9	0.30 mm × 75 mm	1	0.04
10	0.40 mm × 50 mm	1	0.04
11	0.40 mm × 60 mm	1	0.04

TABLE 4 Frequency of retention time.

No.	Retention time (min)	Frequency	Percentage
1	30	24	0.71
2	20	5	0.15
3	10	2	0.06
4	40	2	0.06
5	5	1	0.03

3.8 Association rule mining analysis

Association rule analysis of the Apriori algorithm was performed using R software (version 4.3.0) to obtain 72 association rules. A grouping matrix plot was used to visualize the 72 association rules (Figure 3B). Darker purple circles indicate higher degrees of lift, whereas larger circles indicate higher support.

Based on the minimum support threshold of 20%, the minimum confidence threshold of 90%, and an uplift factor of>1, 20 pairs of acupoint combinations with the highest support thresholds in the PSD prescriptions were summarized. The first 12 pairs included {EX-HN12} ≧ {EX-HN13}, {EX-HN13} ≧ {EX-HN12}, {EX-HN12} ≧ {GB20}, {EX-HN13} ≧ {GB20}, {EX-HN12, EX-HN13} ≧ {GB20}, {EX-HN12, GB20} ≧ {EX- HN13}, {EX-HN13, GB20} ≧ {EX-HN12}, {EX-HN12, RN23} ≧ {EX-HN13}, {EX-HN13, RN23} ≧ {EX-HN12}, {GB12} ≧ {GB20}, {Zhiqiang} ≧ {Tunyan}, and {SJ17} ≧ {GB20}. The top 20 pairs of acupoint combinations had confidence, support, and lift values (Table 11), and more than half of the acupoint combinations had confidence values of 100%. Notably, all 20 pairs of acupoint combinations had lift values >1, with nine acupoint combinations >2.

TABLE 5 Frequency of treatment.

No.	Frequency of treatment	Frequency	Percentage
1	5 days per week	15	0.39
2	6 days per week	12	0.32
3	7 days per week	10	0.26
4	3 days per week	1	0.03

TABLE 6 Frequency of total treatment duration.

No.	Total treatment duration	Frequency	Percentage
1	3 weeks	22	0.55
2	2 weeks	6	0.15
3	3 weeks	6	0.15
4	8 weeks	4	0.10
5	6 weeks	2	0.05

3.9 Complex network analysis

A complex network analysis was performed using Cytoscape to derive 38 acupoints (nodes) and 210 edges, which were classified into four layers by degree scores: the darker the color and the higher the frequency. Hub acupoints were obtained using the maximal clique centrality (MCC), maximum neighborhood component (MNC), and molecular complex detection tool (MCODE). Specifically, the top 10 acupoints and 45 edges were obtained using MCC in the Cytoscape tool, with the acupoints in descending order as follows: GB20, RN23, EX-HN12, EX-HN13, Tunyan, EX-HN14, Zhiqiang, Gongxue, MS6, and DU20. The top 10 acupoints and 42 edges were obtained using MNCs, including GB20, RN23, EX-HN12, EX-HN13, Tunyan, SJ17, DU16, EX-HN14, MS6, and DU20. Additionally, nine acupoints and 32 edges were obtained using MCODE, including DU20, Fayin, Tunyan, EX-HN14, Zhiqiang, Wai EX-HN12, Gongxue, Wai EX-HN13, and Tiyan. Notably, these hub acupoints were consistent with the results of association rule analysis (Figure 4).

3.10 Correlation and cluster analysis

The 20 most frequently used acupoints were clustered and correlated into six clusters. Cluster 1 includes EX-HN14, Wai EX-HN12, Wai EX-HN13, Gongxue, Tunyan, and Zhiqiang. Cluster

TABLE 7 Frequency of the top 15 acupoints used for PSD.

No.	Acupoint	Frequency	Meridians	Site of the point	Specific points
1	GB20	34	GB	Head, face, and neck	Crossing point
2	RN23	31	RN	Head, face, and neck	Crossing point
3	EX-HN12	14	EX	Head, face, and neck	NA
4	EX-HN13	14	EX	Head, face, and neck	NA
5	Tunyan	13	Other	Head, face, and neck	NA
6	SJ17	12	SJ	Head, face, and neck	Crossing point
7	EX-HN14	11	EX	Head, face, and neck	NA
8	GB12	11	GB	Head, face, and neck	NA
9	Gongxue	11	Other	Head, face, and neck	NA
10	Zhiqiang	11	Other	Head, face, and neck	NA
11	DU16	12	DU	Head, face, and neck	Crossing point
12	MS6	10	MS	Head, face, and neck	NA
13	DU20	9	DU	Head, face, and neck	Crossing point
14	Wai EX-HN12	9	Other	Head, face, and neck	NA
15	Wai EX-HN13	9	Other	Head, face, and neck	NA

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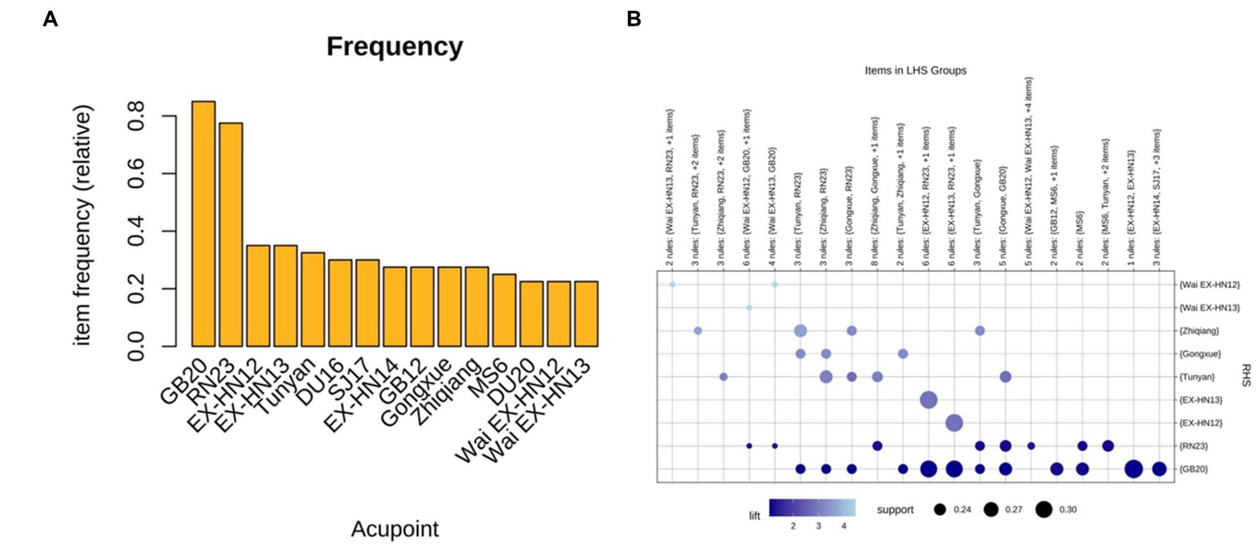

FIGURE 3
(A) Frequency of the top 15 acupoints used for PSD. **(B)** Grouping matrix of 72 association rules for acupuncture intervention PSD.

TABLE 8 Frequency and percentage of meridians used for PSD.

Meridians	Frequency	Percentage	Acupoint number	Acupoints (frequency)
Other	80	0.25	19	Tunyan (13), Gongxue (11), Zhiqiang (11), Wai EX-HN12 (9), Wai EX-HN13 (9), Pang RN23 (5), Fayin (4), Shang RN23 (4), Yanhoubi (3), Tiyan (2), Aqiang (1), Ganqu (1), Piqu (1), Shangjiaoqu (1), Shezhong (1), Siqiang (1), Tunyan2 (1), Xinqu (1), Zhifanliu (1)
GB	45	0.14	2	GB20 (34), GB12 (11)
EX	43	0.13	6	EX-HN12 (14), EX-HN13 (14), EX-HN14 (11), EX-B2 (2), EX-HN11 (1), EX-HN5 (1)
RN	43	0.13	6	RN23 (31), RN22 (7), RN24 (2), RN12 (1), RN4 (1), RN6 (1)
DU	31	0.10	7	DU16 (12), DU20 (9), DU26 (4), DU15 (3), DU17 (1), DU24 (1), DU29 (1)
MS	20	0.06	5	MS6 (10), MS7 (6), MS5 (2), MS1 (1), MS9 (1)
SJ	13	0.04	2	SJ17 (12), SJ5 (1)
ST	12	0.04	6	ST9 (5), ST36 (2), ST40 (2), ST25 (1), ST4 (1), ST6 (1)
LI	9	0.03	4	LI4 (6), LI11 (1), LI14 (1), LI18 (1)
BL	6	0.02	5	BL10 (2), BL13 (1), BL18 (1), BL20 (1), BL23 (1)
PC	5	0.02	1	PC6 (5)
SP	5	0.02	2	SP6 (4), SP4 (1)
KI	4	0.01	3	KI6 (2), KI1 (1), KI3 (1)
LU	2	0.01	1	LU7 (2)
LR	2	0.01	1	LR3 (2)
HT	2	0.01	1	HT5 (2)

GB, Gallbladder Meridian of Foot Shaoyang; EX, Extra points Meridian; RN, Conception Vessel; DU, Governor Vessel; MS, Anterior parietotemporal oblique line, SJ, San Jiao Meridian of Hand Shaoyang; ST, Stomach Meridian of Foot Yangming; LI, Large Intestine Meridian of Hand Yangming; BL, the Bladder Meridian of Foot Taiyang; PC, Pericardium Meridian of Hand Jueyin; SP, Spleen Meridian of Foot Taiyin; KI, Kidney Meridian of Foot Shaoyin; LU, Lung Meridian of Hand Taiyin; LR, Liver Meridian of Foot Jueyin; HT, Heart Meridian of Hand Shaoyin.

2 comprised EX-HN12 and EX-HN13. Cluster 3 included strains SJ17 and GB12. Cluster 4 includes RN23, GB20, MS6, and MS7. Cluster 5 included DU20, Pang RN23, and RN22. Cluster 6 included DU16, LI4, and PC6 (Figure 5).

4 Discussion

We organized and established a database of unstructured data from studies of acupuncture intervention in PSD using data mining

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TABLE 9 Frequency and percentage of specific acupoints used for PSD.

Specific acupoints	Frequency	Percentage	Acupoint number	Acupoints (frequency)
Crossing point	123	0.38	15	GB20 (34), RN23 (31), SJ17 (12), DU16 (12), DU20 (9), RN22 (7), DU26 (4), SP6 (4), DU15 (3), RN24 (2), DU17 (1), DU24 (1), RN12 (1), RN4 (1), ST4 (1)
Luo-connecting point	13	0.04	6	PC6 (5), HT5 (2), LU7 (2), ST40 (2), SJ5 (1), SP4 (1)
Eight confluent point	11	0.03	5	PC6 (5), KI6 (2), LU7 (2), SJ5 (1), SP4 (1)
Yuan-primary point	9	0.03	3	LI4 (6), LR3 (2), KI3 (1)
Five Shu points	7	0.02	5	LR3 (2), ST36 (2), KI1 (1), KI3 (1), LI11 (1)
Back Shu point	4	0.01	4	BL13 (1), BL18 (1), BL20 (1), BL23 (1)
Front Mu point	3	0.01	3	RN12 (1), RN4 (1), ST25 (1)
Lower He-Sea point	3	0.01	2	ST36 (2), LI11 (1)
Eight influential point	1	0.00	1	RN12 (1)

TABLE 10 Frequency and percentage of acupoint sites used for PSD.

Site of acupoints	Frequency	Percentage	Acupoint number	Acupoints (frequency)
Head, face, and neck	279	0.87	47	GB20 (34), RN23 (31), EX-HN12 (14), EX-HN13 (14), Tunyan (13), SJ17 (12), DU16 (12), EX-HN14 (11), GB12 (11), Gongxue (11), Zhiqiang (11), MS6 (10), DU20 (9), Wai EX-HN12 (9), Wai EX-HN13 (9), RN22 (7), MS7 (6), Pang RN23 (5), ST9 (5), DU26 (4), Fayin (4), Shang RN23 (4), DU15 (3), Yanhoubi (3), EX-B2 (2), MS5 (2), RN24 (2), Tiyan (2), Aqiang (1), DU17 (1), DU24 (1), DU29 (1), EX-HN10 (1), EX-HN5 (1), Ganqu (1), LI18 (1), MS1 (1), MS9 (1), Piqu (1), Shangjiaoqu (1), Shezhong (1), Siqiang (1), ST4 (1), ST6 (1), Tunyan2 (1), Xinqu (1), Zhifanliu (1)
Upper limbs	18	0.06	7	LI4 (6), PC6 (5), HT5 (2), LU7 (2), LI11 (1), LI14 (1), SJ5 (1)
Lower limbs	15	0.05	8	SP6 (4), KI6 (2), LR3 (2), ST36 (2), ST40 (2), KI1 (1), KI3 (1), SP4 (1)
Back	6	0.02	5	BL10 (2), BL13 (1), BL18 (1), BL20 (1), BL23 (1)
Chest and abdomen	4	0.01	4	RN12 (1), RN4 (1), RN6 (1), ST25 (1)

techniques to summarize the parameters related to acupuncture in PSD over the past 15 years and to provide evidence-based guidance for future clinical data studies and methodological research. Specifically, we integrated 39 RCTs of acupuncture intervention for PSD and extracted needling parameters (needling type, retention time, treatment frequency, and treatment period) and acupoint parameters (frequency of acupoints, meridians, specific acupoints, and acupoint sites) from the studies with positive outcomes. Moreover, the correlation rules, complex networks, clustering, and correlations of the acupoints were analyzed. These results have positive implications for acupuncturists when making prescription decisions for the treatment of PSD.

Regarding needling parameter patterns, we identified the two most frequently used needling types, 0.25 mm × 40 mm and 0.30 mm × 40 mm, and the optimal retention time of 30 min. Clinical acupuncturists varied somewhat in their choice of needle type and retention time owing to inconsistent practice backgrounds. However, our results may indicate an overall trend in acupuncture parameters for PSD. Although the frequency of treatment suggests that five times per week is optimal, recent studies have demonstrated different after-effects of acupuncture at different intervals (50). Specifically, short intervals (6 h) somewhat blocked acupuncture after the effects, and long intervals (48 h) prolonged them. Regarding the total intervention, a four-week intervention may be optimal for the recovery of patients with PSD. This result may depend on inconsistent point selection criteria by acupuncturists, and the efficacy may be affected by confounding factors.

Regarding acupoint parameters, the commonly used acupoints were GB20 and RN23. Internally, it passes through the branches of the greater occipital nerve, the lesser occipital nerve, and the occipital artery, and it is innervated by the vagus nerve and glossopharyngeal nerve. Acupuncture of the GB20 stimulates the neck muscles, improves blood supply to the vertebrobasilar artery system, increases blood flow to the brain, promotes nerve repair and regeneration, and restores swallowing strength in patients with PSD (51, 52). Anatomically, RN23 is surrounded by the hyoid muscle, genioglossus muscle,

TABLE 11 The top 20 acupoint association rules for PSD treatments.

No.	Association rules	Support	Confidence	Lift
1	{EX-HN12} ≧ {EX-HN13}	0.35	1.00	2.86
2	{EX-HN13} ≧ {EX-HN12}	0.35	1.00	2.86
3	{EX-HN12} ≧ {GB20}	0.33	0.93	1.09
4	{EX-HN13} ≧ {GB20}	0.33	0.93	1.09
5	{EX-HN12, EX-HN13} ≧ {GB20}	0.33	0.93	1.09
6	{EX-HN12, GB20} ≧ {EX-HN13}	0.33	1.00	2.86
7	{EX-HN13, GB20} ≧ {EX-HN12}	0.33	1.00	2.86
8	{EX-HN12, RN23} ≧ {EX-HN13}	0.30	1.00	2.86
9	{EX-HN13, RN23} ≧ {EX-HN12}	0.30	1.00	2.86
10	{GB12} ≧ {GB20}	0.28	1.00	1.18
11	{Zhiqiang} ≧ {Tunyan}	0.28	1.00	3.08
12	{SJ17} ≧ {GB20}	0.28	0.92	1.08
13	{EX-HN12, RN23} ≧ {GB20}	0.28	0.92	1.08
14	{EX-HN13, RN23} ≧ {GB20}	0.28	0.92	1.08
15	{EX-HN12, EX-HN13, RN23} ≧ {GB20}	0.28	0.92	1.08
16	{EX-HN12, GB20, RN23} ≧ {EX-HN13}	0.28	1.00	2.86
17	{EX-HN13, GB20, RN23} ≧ {EX-HN12}	0.28	1.00	2.86
18	{MS6} ≧ {GB20}	0.25	1.00	1.18
19	{EX-HN14} ≧ {GB20}	0.25	0.91	1.07
20	{Zhiqiang} ≧ {RN23}	0.25	0.91	1.17

glossopharyngeal nerve, and hypoglossal nerves. Acupuncture stimulates local nerve points, rebuilds the reflex arc of the central nervous system excitability, restores pharyngeal innervation, and completes involuntary movement (53, 54). Moreover, the GB, EX, RN, and nontraditional meridians are most frequently used in the clinical treatment of PSD. These meridians pass through the pharynx, and needling acupoints on the meridians reflect the traditional medical principle of "wherever the meridians pass, the main treatment will be applied." Joint regulation of multiple meridians promotes blood circulation in the glossopharynx and restores normal swallowing function. Compared to the lower He-Sea point, five Shu points, and eight influential points, the crossing point is a more important acupoint in PSD selection. For example, GB20 belongs to the GB meridian, which is the meeting point for the GB, Sanjiao, and Yangwei meridians. Acupuncture of the GB meridian can simultaneously regulate all three meridians, which play a role in the recovery of patients with PSD.

Our findings indicate that the acupoints are mostly located on the head, face, and neck, which is consistent with the etiology of dysphagia (mostly cerebrovascular disease) and the location of lesions (located in the cerebral cortex or subcortex). Thus, needling the head acupoints stimulates the corresponding swallowing function area of the brain, which can improve cerebral blood flow, regulate growth factors, reduce inflammatory factors, and promote the recovery of neuronal cells in the lesion location of the brain (55). Additionally, needling peripheral areas, such as the face and neck, stimulate multiple brain nerves that innervate swallowing movements, such as the trigeminal, facial, glossopharyngeal, vagus, and hypoglossal nerves. These stimulation signals are transmitted to the swallowing center and cerebral cortex, which promote compensatory brain function and recovery of the swallowing muscles. Furthermore, acupoint association rule analysis identified the most strongly associated core acupoints: GB20, RN23, EX-HN14, Gongxue, MS6, SJ17, EX-HN12, and EX-HN13. Data mining analysis using frequency counts, clustering, and correlation indicated that the acupoints with the strongest evidence of acupuncture in the PSD group were GB20, RN23, and MS6. Our study provides more choices of parameters for acupuncture in PSD, and the specificity of acupoints should be emphasized based on the effectiveness obtained by acupuncturists in clinical practice.

Qiao and colleagues measured the characteristics of dysphagia in individuals with stroke at different lesion locations through the duration, movement, and swallowing function using Videofluoroscopic Swallow Study (56). The report indicates that patients with infratentorial strokes had worse swallowing functions compared to those with supratentorial strokes. However, a single-center retrospective study showed that the risk of PSD was reduced in patients receiving acupuncture treatment, regardless of whether the stroke occurred in the brainstem [adjusted hazard ratio (AHR) = 0.41], thalamus (AHR = 0.13), or was a multifocal lesion (AHR = 0.40) (57). Although the results indicate that acupuncture treatment can reduce the risk of dysphagia in stroke patients across various demographics, including different ages, sexes, stroke types, sites, and baseline comorbidities (57), we recognize the need to further elucidate the application of acupuncture techniques in diverse clinical settings. Additionally, our standardized treatment protocols require further evaluation in future research to determine their effectiveness and applicability.

This study had some limitations that should be interpreted with caution by clinicians and researchers. First, 27 studies reported the

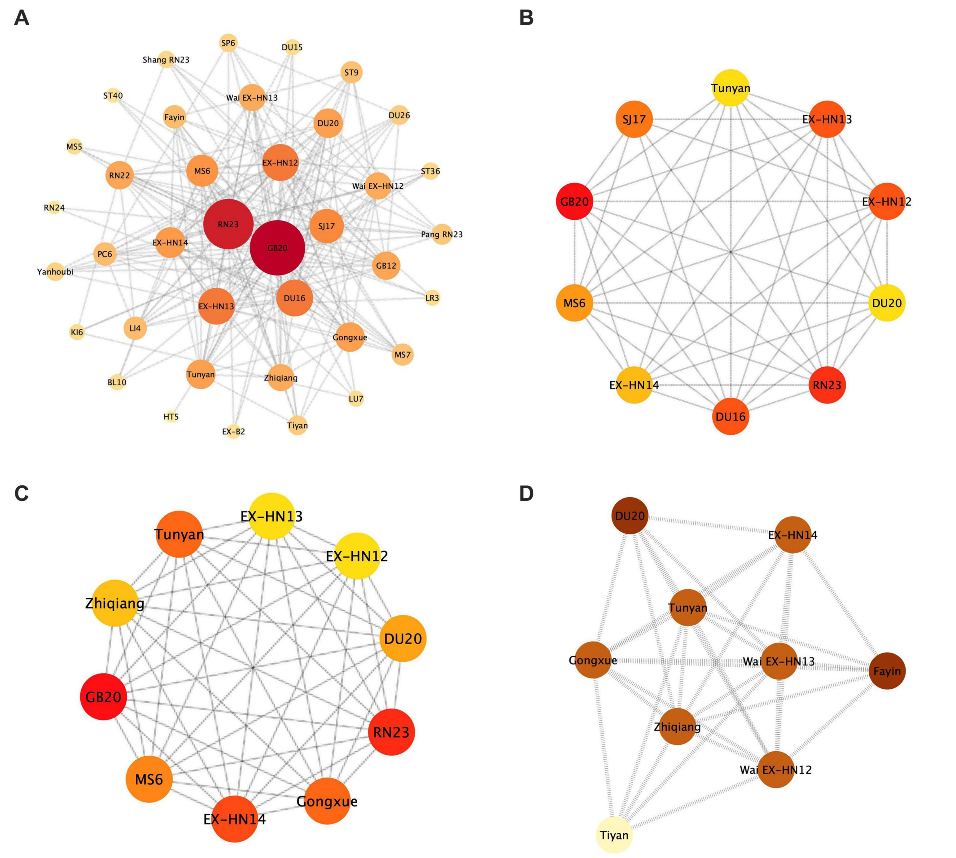

FIGURE 4
Complex network analysis. **(A)** Complex network analysis of acupuncture for PSD treatment. **(B)** MNC shows the top 10 acupuncture points. **(C)** MCC shows the top 10 acupuncture points. **(D)** MCODE shows 9 acupoints and 32 edges.

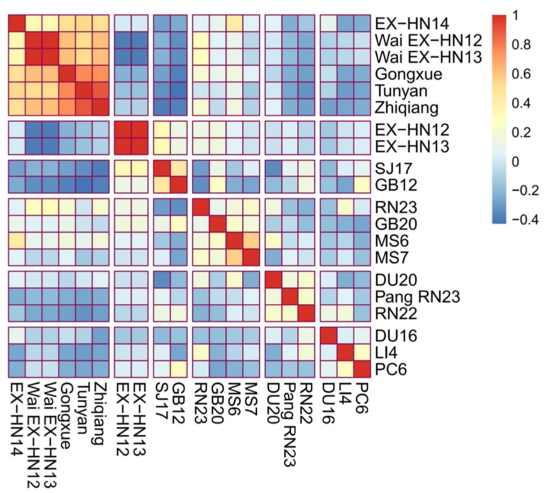

FIGURE 5
Correlation and cluster analysis of the top 20 frequently selected acupoints.

WST as an outcome indicator; however, the guidelines for PSD suggest that the WST should be used for initial screening. Instrumentation (FESS and VFSS), combined with scale interpretation as an outcome assessment, can objectively confirm the degree of recovery from dysphagia. Second, most studies did not report the practice of acupuncturists, and untrained or inexperienced acupuncturists may result in different therapeutic and physiological outcomes. Future studies should follow the STRICTA guidelines to improve the quality of the evidence. Third, the 39 RCTs included poorer methodological quality, lack of allocation concealment, and inability to confirm preregistered protocols, which would carry some risk of bias. Finally, we performed strong data integration of acupuncture type, treatment duration, and acupoint association. However, further large-sample RCTs or animal experiments are needed to validate our results.

5 Conclusion

This study utilized data mining methods to summarize the optimal parameters and clinical acupuncture point selection characteristics for acupuncture treatment of PSD. The closest parameter combination included needle sizes of 0.25 mm × 40 mm, a needle retention time of 30 min, treatment frequency of five times per week, and a total treatment course of four weeks. Additionally, the core points identified were GB20, RN23, EX-HN14, Gongxue, MS6, SJ17, EX-HN12, and EX-HN13, with the principal combinations being EX-HN12, EX-HN13, GB20, and RN23. Due to the limitations of this study, further research and more standardized clinical trials are still needed to guide and optimize acupuncture treatment plans, providing a theoretical basis for clinical acupuncturists.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

MW: Conceptualization, Data curation, Software, Writing – original draft, Writing – review & editing. WS: Formal analysis, Investigation, Methodology, Writing – review & editing. XW: Investigation, Methodology, Visualization, Writing – review & editing. QT: Conceptualization, Supervision, Validation, Writing – review & editing. WG: Supervision, Validation, Writing – review & editing. LZ: Conceptualization, Funding acquisition, Project administration, Supervision, Validation, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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A systematic review and meta-analysis of acupuncture in aspiration caused by post-stroke dysphagia

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Objective: This systematic review and meta-analysis aims to systematically evaluate the effectiveness and safety of acupuncture in the treatment of aspiration caused by post-stroke dysphagia.

Methods: A computer search was conducted in nine databases, including the China National Knowledge Infrastructure (CNKI), China Science and Technology Journal (VIP), Wan-fang Database, China Biomedical Literature Database (CBM), PubMed, Web of Science, Cochrane Library, Embase, and Chinese Clinical Trial Registry (ChiCTR), from their inception until April 2024. Clinical randomized controlled trials comparing acupuncture combined therapy or single therapy with control interventions for the treatment of aspiration caused by post-stroke dysphagia were included. The primary outcome measure was the Penetration Aspiration Scale (PAS), and secondary outcome measures included the overall effective rate, video fluoroscopic swallowing study (VFSS), and hyoid bone displacement. The statistical analysis was performed using RevMan 5.3 and Stata 16.0.

Results: A total of 16 articles involving 1,284 patients were included. The meta-analysis results showed that acupuncture combined therapy or single therapy was more effective in improving PAS scores compared to conventional rehabilitation therapy or balloon dilation of the catheter [WMD = –1.05, 95% CI (–1.30, –0.80), $Z = 0.82$, $p = 0.00 < 0.05$]. It was also more effective in improving VFSS scores [WMD = 1.32, 95% CI (0.08, 2.55), $Z = 2.09$, $p = 0.04 < 0.05$] and hyoid bone displacement [WMD = 2.02, 95% CI (0.86, 3.18), $Z = 3.41$, $p = 0.00 < 0.05$]. Additionally, acupuncture had a higher overall effective rate [WMD = 1.21, 95% CI (1.14, 1.29), $Z = 5.76$, $p = 0.00 < 0.05$] and a lower incidence of adverse events. Sensitivity analysis indicated that the literature had minimal impact on the results, and bias tests showed no publication bias.

Conclusion: Acupuncture combined therapy and acupuncture single therapy can effectively improve aspiration caused by post-stroke dysphagia with a low incidence of adverse events. However, due to the low quality of the included literature, more high-quality randomized controlled trials are still needed to confirm the effectiveness and safety of acupuncture in the treatment of aspiration caused by post-stroke dysphagia.

Systematic review registration: https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42023462707, identifier CRD42023462707

KEYWORDS

acupuncture, stroke, dysphagia, aspiration, systematic review, meta-analysis

1 Introduction

Aspiration refers to the phenomenon of the entry of oral or gastric contents below the glottis into the respiratory tract. It is the most severe and immediate consequence of post-stroke dysphagia (PSD) (1) and one of the leading factors causing pneumonia (2). Following aspiration, patients immediately experience irritative coughing, dyspnea, and even asthma, known as dominant aspiration (DA). Silent aspiration (SA) refers to the absence of external signs such as coughing within 1 min after aspiration, without symptoms of irritative coughing or dyspnea. The incidence of aspiration in PSD ranges from 15 to 54% (2, 3), with approximately 68% being SA, which is difficult to detect through non-imaging examinations (3, 4), leading to potential underdiagnosis. Hence, the actual incidence of SA may be higher (5). Aspiration can result in malnutrition, pneumonia, and even suffocation (6). Studies have shown that the occurrence rate of pneumonia in patients with aspiration is 11 times higher than in other patients (7), and the overall 30 day mortality rate of aspiration pneumonia is 21–30% (8), significantly affecting patient prognosis, prolonging hospital stays, and increasing treatment costs (9).

Clinical guidelines in the United States (10), the United Kingdom (11), Germany (12), and other countries recommend that acute stroke patients should undergo swallowing screening by trained healthcare professionals within 4 h of hospital admission or before taking any solid, liquid medications, or food. Many screening criteria include the presence of coughing symptoms (5). However, SA is more covert clinically, as patients may not exhibit symptoms such as coughing. Therefore, SA cannot be easily determined through clinical swallowing screening and requires imaging examinations. Currently, video fluoroscopic swallowing study (VFSS) is considered the gold standard for diagnosing aspiration both domestically and internationally (13, 14). It utilizes natural eating methods to create visual images of tongue movement, food transmission in the pharynx, elevation of the hyoid bone and larynx, and movement of the soft palate and epiglottis. By observing the flow of contrast agent from the oral cavity to the cervical esophagus and determining the depth of contrast agent entry into the airway, penetration or aspiration can be accurately assessed, reducing missed diagnoses (15). The Penetration Aspiration Scale (PAS) is an assessment scale derived from VFSS, which allows clinicians to visually understand the presence of aspiration in stroke patients through numerical scores. It provides a reliable quantitative indicator for evaluating the severity of aspiration and serves as a reliable tool for swallowing function assessment (16).

The mechanism of aspiration caused by PSD remains unclear and may be related to weakened pharyngeal reflexes, weakened pharyngeal muscle strength, and decreased coordination due to nerve damage. It could also be associated with reduced pharyngeal sensation, impaired reflex coughing ability, or low levels of P substance and dopamine (3). Modern medical treatments for post-stroke dysphagia and aspiration include swallowing training, local muscle stimulation, dietary improvements, and changes in eating positions. Although these methods have proven effectiveness, they require evaluation by specialized rehabilitation therapists, and their efficacy may vary among different populations (1). Therefore, the discovery of safe and effective treatment methods holds immense clinical significance.

Acupuncture, as a simple, environmentally friendly, and cost-effective treatment method, has been supported by a large number of clinical studies and evidence-based medicine to effectively treat post-stroke dysphagia (17, 18). The Chinese Expert Consensus on Swallowing Disorders and Nutritional Management in Stroke Patients also recommends acupuncture as a Class A treatment method (19). However, some studies on aspiration caused by PSD have used non-imaging examinations as outcome measures (20), or the outcome measures lacked a quantitative scale for analyzing the severity of aspiration (21), which cannot demonstrate the efficacy of acupuncture for SA. Therefore, this study conducted a meta-analysis to evaluate the efficacy and safety of acupuncture in the treatment of aspiration caused by post-stroke dysphagia, aiming to provide evidence-based medicine support for its effectiveness.

2 Methods

This study has been registered in PROSPERO (registration number: CRD42023462707), and the registration details can be obtained from https://www.crd.york.ac.uk/prospero/. The reporting of study results follows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) 2020 checklist (see Appendix 1).

2.1 Data sources and search strategy

A computer search was conducted in nine databases, including CNKI, VIP, WF, SinoMed, PubMed, Web of Science, Cochrane Library, EMbase, and ChiCTR, from their inception until April 2024. Chinese subject terms and free terms were determined using the subject term search function in SinoMed, while English subject terms and free terms were determined using the MeSH Database in PubMed. The Chinese subject terms included "卒中" (stroke), "吞咽障碍" (deglutition disorders), "针刺" (acupuncture), and "随机对照试验" (randomized controlled trial). The English subject terms included "stroke," "Deglutition Disorders," "acupuncture," and "randomized controlled trial." There were no language restrictions on the literature search. The specific search terms and strategies are detailed in Appendix 2.

2.2 Inclusion criteria

1. Study type: randomized controlled trials.
2. Study population: patients diagnosed with stroke according to the guidelines for the prevention of stroke issued by the American Heart Association and the American Stroke Association (22) or the diagnostic criteria for stroke in the Chinese Guidelines for the Prevention and Treatment of Stroke (2021 edition) (23). The patients should also meet the diagnostic criteria for swallowing disorders established by the European Stroke Organization and the European Society for Swallowing Disorders (24).
3. Intervention: the experimental group received acupuncture therapy alone or in combination with other therapies compared to the control group, which received any therapy other than acupuncture.
4. Primary outcome measure: Rosenbek Penetration Aspiration Scale (PAS). Secondary outcome measures included overall effective

rate, video fluoroscopic swallowing study (VFSS), and hyoid bone displacement. The studies had to include the primary outcome measure and at least one of the secondary outcome measures.

2.3 Exclusion criteria

1. Subjects with other conditions affecting swallowing function, such as tumors, myasthenia gravis, Parkinson's syndrome, Guillain-Barré syndrome, etc.
2. Subjects with other life-threatening diseases, such as severe coronary heart disease, heart failure, severe pneumonia, etc.
3. Duplicate publications.
4. Literature with incomplete trial data.
5. Literature for which the full text could not be obtained.

2.4 Literature screening and data extraction

Following the methods for including studies described in the Cochrane Handbook for Systematic Reviews of Interventions (25), the search results from each database were imported into the reference management software NoteExpress. The screening process was conducted based on the inclusion and exclusion criteria, as shown in Figure 1. The eligible literature was then organized and recorded in Excel, including information such as study authors, publication dates, sample sizes, gender distribution, average age, intervention measures, treatment frequency and duration, outcome measures, and adverse events. Two independent reviewers conducted the literature screening and data extraction, with cross-checking of the results. If any discrepancies arose during the study selection or data extraction process, a third reviewer intervened to make a judgment.

When outcome measures were presented as medians (m), first quartile ($q1$), third quartile ($q3$), and/or minimum (a) and maximum (b) values, they were transformed into mean ± standard deviation using statistical conversion formulas (26, 27).

$$S \approx \frac{b-a}{2\Phi^{-1}\left(\frac{n-0.375}{n+0.25}\right)} \quad S \approx \frac{q_3-q_1}{2\Phi^{-1}\left(\frac{0.75n-0.125}{n+0.25}\right)}$$

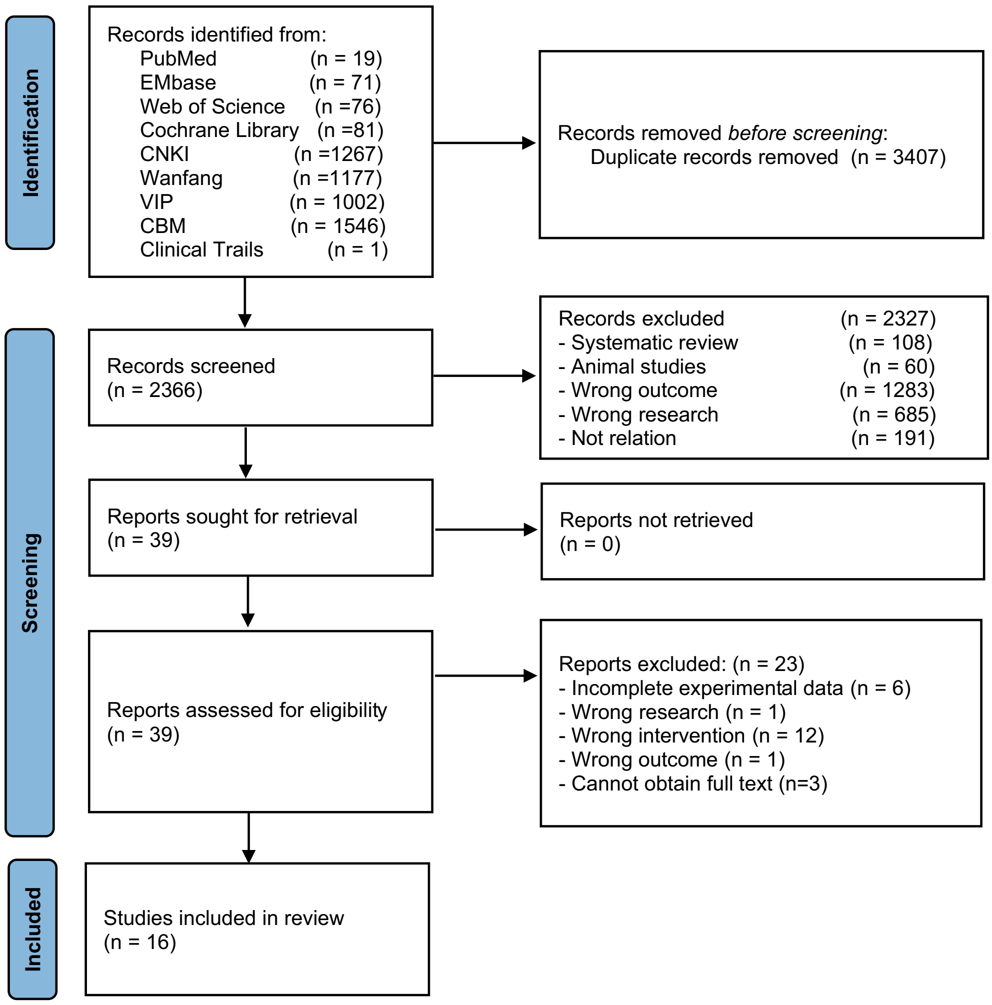

FIGURE 1
Flowchart of the literature search process for acupuncture treatment of aspiration caused by post-stroke dysphagia.

$$\bar{X}(w) \approx \left(\frac{4}{4+n^{0.75}}\right)\frac{a+b}{2} + \left(\frac{n^{0.75}}{4+n^{0.75}}\right)m$$

$$\bar{X}(w) \approx \left(0.7+\frac{0.39}{n}\right)\frac{q_1+q_3}{2} + \left(0.3-\frac{0.39}{n}\right)m$$

When an outcome measure is presented in the form of multiple subgroups, the data from the subgroups are combined using the formula provided in the Cochrane Handbook for Systematic Reviews of Interventions (28).

$$\text{Sample size}\left(\text{Combined groups}\right) = N1 + N2.$$

$$\text{Mean}\left(\text{Combined groups}\right) = \frac{N_1 M_1 + N_2 M_2}{N_1 + N_2}$$

$$\text{SD}\left(\text{Combined groups}\right) = \sqrt{\frac{(N_1-1)\text{SD}_1^2 + (N_2-1)\text{SD}_2^2 + \frac{N_1 N_2}{N_1+N_2}\left(M_1^2 + M_2^2 - 2M_1M_2\right)}{N_1+N_2-1}}$$

2.5 Quality assessment

All studies included in this research were randomized controlled trials. Therefore, the Risk of Bias 2 (RoB 2) tool provided by the Cochrane Collaboration, as recommended by the GRADE evidence grading system, was utilized to assess the risk of bias in the included studies (29). The assessment criteria included random sequence generation, allocation concealment, blinding, completeness of outcome data, selective reporting, and other biases. The included literature was categorized as having low, unclear, or high risk of bias based on the assessment criteria. Two trained reviewers (JL and XW) independently assessed the included studies using RoB 2 and calculated the intra-class correlation coefficient (ICC) to conduct a consistency test. If the consistency reached at least 80%, a formal assessment was conducted. Any discrepancies were resolved through discussion with a third reviewer (ZZ).

2.6 Statistical analysis

Statistical analysis was conducted using RevMan 5.3 and Stata 16.0. The analysis included tests for baseline consistency, heterogeneity, sensitivity analysis, and publication bias assessment using funnel plots when the number of included studies exceeded 5. For continuous variables, mean differences (MD) were used as the effect size, and for binary variables, risk ratios (RR) were used as the effect size. The 95% confidence intervals (CI) were calculated, and a p-value <0.05 was considered statistically significant. The criteria for heterogeneity testing were based on the Cochrane Handbook for Systematic Reviews of Interventions 5.0.2 (30). Heterogeneity was assessed by calculating the I^2 statistic and the p-value from the Q test. I^2 values between 0 and 40% indicated low heterogeneity, 30 to 60% indicated moderate heterogeneity, 50 to 90% indicated substantial heterogeneity, and $I^2 > 75\%$ indicated high heterogeneity. Additionally, a p-value <0.1 from the Q test indicated significant heterogeneity. In this study, a fixed-effects model was used for analysis when $I^2 \leq 50\%$ and $p \geq 0.1$, and a random-effects model was used for analysis when $I^2 > 50\%$ and $p < 0.1$. Meta-regression was conducted to identify the sources of heterogeneity. Sensitivity analysis was performed using the one-by-one exclusion method, and if the pooled effect size fell outside the 95% confidence interval, it was considered to have a strong impact on the study results. Bias assessment was conducted by plotting a funnel plot and calculating the bias p-value, where a p-value >0.05 indicated no publication bias.

3 Results

3.1 Literature selection

A total of 5,773 articles were identified through the database search in nine databases (CNKI = 1,267, VIP = 1,002, WF = 1,711, CBM = 1,546, PubMed = 19, Web of Science = 76, Cochrane Library = 81, Embase = 71, Clinical Trials = 1). After removing duplicates (n = 3,407), 2,327 articles were excluded based on the title and abstract. Full-text screening resulted in the exclusion of 23 articles, and finally, 16 articles were included (31–46). The flowchart of the literature selection process is shown in Figure 1.

3.2 Characteristics of included studies

The 16 included studies were all single-center randomized controlled trials conducted in China, involving a total of 1,284 cases, with 642 cases in the experimental group and 642 cases in the control group. Three studies were master's theses (40, 42, 45), and the remaining 13 were journal articles. The average age of the patients ranged from 56 to 71 years. The average duration of stroke was less than 1 day in one study (36), more than 3 months in one study (44), and unreported in one study (41), while the average duration in the remaining studies ranged from 15 days to 75 days. The proportion of stroke types was not reported in two studies (39, 41), one study included only ischemic stroke patients (37), and the remaining studies included more ischemic stroke patients than hemorrhagic stroke patients. Three studies used acupuncture combined therapy (41, 43, 46), while the remaining studies used acupuncture monotherapy. One study used electroacupuncture (35), one study used auricular acupressure (33), and the remaining studies used conventional acupuncture. All studies compared the PAS scores, nine studies compared the overall effective rate (31–33, 36, 39, 40, 42, 44, 46), four studies compared VFSS scores (36, 41, 42, 44), and three studies compared hyoid bone displacement (38, 40, 43). The basic characteristics of the included studies are presented in Table 1.

3.3 Risk of bias in the included studies

The ICC value for the RoB 2 assessment by the two reviewers was 0.848, indicating excellent consistency.

TABLE 1 Basic characteristics of included studies on acupuncture treatment for aspiration caused by post-stroke dysphagia.

Author and year of publication	Course	Frequency of treatment	Adverse events		Type of stroke (Cases)		Interventions	Course (Days)	Age (Years)	Finale index
					Haemorrhage	Ischemia				
Cao 2023 (31)	4 weeks	6 times a week	Not mentioned	T	13	17	Acupuncture	34.0 ± 22.1	61 ± 8	PAS, Overall effective rate
				C	12	18	NMES	36.8 ± 47.4	59 ± 7	
Chen 2018 (32)	4 weeks	5 times a week	Not mentioned	T	9	21	Acupuncture	39.20 ± 12.61	62.90 ± 10.04	PAS, Overall effective rate
				C	6	24	Swallowing training	36.83 ± 14.6	63.00 ± 9.83	
Chen 2022 (33)	2 weeks	7 times a week	Not mentioned	T	11	29	Auriculopathic compression	62.05 ± 7.08	62.05 ± 7.08	PAS, Overall effective rate
				C	18	22	Swallowing training	59.70 ± 5.80	59.70 ± 5.80	
Huang 2021 (34)	3 weeks	5 times a week	Not mentioned	T	8	13	Acupuncture	64.68 ± 38.71	70.90 ± 7.09	PAS
				C	7	14	NMES	64.68 ± 44.38	71.33 ± 6.39	
Kang 2023 (35)	3 weeks	5 times a week	Not mentioned	T	3	12	Electroacupuncture	26.87 ± 29.57	61.80 ± 12.45	PAS
				C	6	24	NMES+Swallowing training	41.20 ± 33.95	63.44 ± 10.83	
Li 2017 (36)	8 weeks	1 time per day	Not mentioned	T	17	23	Acupuncture	0.35 ± 0.72	63.6 ± 11.34	PAS, FESS, overall effective rate
				C	33	37	Swallowing training	0.37 ± 0.42	63.57 ± 11.08	
Li 2019 (37)	4 weeks	6 times a week	Not mentioned	T	0	40	Acupuncture	16.9 ± 7.1	61.9 ± 7.9	PAS
				C	0	40	Swallowing training	18.5 ± 8.1	63.6 ± 6.9	
Lin 2021 (38)	4 weeks	5 times a week	Not mentioned	T	17	28	Acupuncture	47.50 ± 9.90	60.40 ± 9.00	PAS, Hyoid bone displacement
				C	14	31	Rtms	46.80 ± 9.70	59.70 ± 9.20	
Lin 2022 (39)	4 weeks	5 times a week	None	T	Not mentioned	Not mentioned	Acupuncture	50.1 ± 20.9	61.00 ± 14.00	PAS, Overall effective rate
				C	Not mentioned	Not mentioned	Swallowing training	48.4 ± 24.9	60 ± 13	
Mo 2021 (40)	4 weeks	6 times a week	None	T	16	24	Acupuncture	73.65 ± 6.41	68.28 ± 0.67	PAS, Hyoid bone displacement, Overall effective rate
				C	18	22	BDC	74.73 ± 6.80	66.73 ± 0.58	
Pan 2023 (41)	3 weeks	5 times a week	Not mentioned	T	Not mentioned	Not mentioned	Acupuncture + Mirror therapy	Not mentioned	62.87 ± 5.43	PAS, FESS
				C	Not mentioned	Not mentioned	Mirror therapy	Not mentioned	65.87 ± 6.35	
Wang 2021 (42)	4 weeks	6 times a week	Not mentioned	T	9	22	Acupuncture	41.32 ± 37.01	63.58 ± 10.29	PAS, FESS, Overall effective rate
				C	11	20	Swallowing training	36.06 ± 37.73	63.90 ± 10.19	
Wu 2022 (43)	4 weeks	5 times a week	Not mentioned	T	35	36	Acupuncture + NMES + BDC	38.01 ± 7.84	57.65 ± 5.73	PAS, Hyoid bone displacement
				C	37	34	NMES+BDC	32.27 ± 7.49	56.82 ± 6.33	
Xia 2020 (44)	3 weeks	5 times a week	Not mentioned	T	12	25	Acupuncture	90 ± 48	60.2 ± 12.3	PAS, FESS, Overall effective rate
				C	13	24	Swallowing training	93 ± 48	66.1 ± 10.5	
Yang 2022 (45)	4 weeks	6 times a week	None	T	13	21	Acupuncture	52.4 ± 17.2	65.5 ± 3.60	PAS
				C	15	19	NMES	51.20 ± 16.70	66.60 ± 3.20	
Zhou 2018 (46)	4 weeks	5 times a week	Not mentioned	T	8	20	Acupuncture + DPNS	15 ± 8	66.7 ± 13.6	PAS, Overall effective rate
				C	9	19	DPNS	17 ± 7	67.0 ± 12.9	

T, intervention group; C, control group; BDC, balloon dilatation Catheter; DPNS, deep pharyngeal neuromuscular stimulation; rTMS, high-frequency repetitive transcranial magnetic stimulation; NMES, neuromuscular electrical stimulation.

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All 16 studies included in the analysis were randomized controlled trials. Among them, 10 studies (32–35, 40, 42–46) utilized random number tables for grouping, one study (36) did not specify the method of randomization, and three studies (31, 39, 44) achieved allocation concealment through opaque envelopes. Two studies (40, 45) experienced participant dropout, with explanations provided for the reasons, and the dropout rates were <10%. The baseline data between the two groups remained comparable, thus not affecting the integrity of the study results. Since all studies obtained informed consent from the participants, blinding was not implemented in any of the studies. Overall, 13 studies (32–38, 42–46) were categorized as having a high risk of bias, while 3 studies (31, 39, 40) were categorized as having an unclear risk of bias. The quality assessment results of the literature are summarized in Figures 2, 3.

3.4 Meta-analysis results

3.4.1 Penetration aspiration scale

A total of 16 studies (31–46) involving the PAS scale were included, with a total of 1,284 cases. The baseline consistency analysis showed low heterogeneity in the baseline effects of the PAS scale between the experimental and control groups (I^2 =0%<50%, and $p = 0.82 > 0.1$ from the Q test). Therefore, a fixed-effects model was selected to combine the effect sizes. The combined effect size for the baseline effects was 0.08 ($z = 1.73$, $p = 0.08 > 0.05$), indicating no statistically significant difference in the baseline period. Subsequent meta-analysis could be performed (see Supplementary Figure S1).

The analysis was stratified into two subgroups based on treatment duration: ≥4 weeks and <4 weeks. There was no statistically significant difference between the subgroups ($p = 0.44$, >0.05, I^2 =0%). Furthermore, even after subgroup analysis, there was still significant heterogeneity in the PAS scale scores ($I^2 = 80 > 50\%$, and the p value of the Q test was $0.000 < 0.05$). Therefore, a random-effects model was chosen for the meta-analysis (see Figure 4).

Due to the high heterogeneity in the combined effect size, meta-regression was conducted to analyze the sources of heterogeneity based on factors such as publication year, intervention measures (acupuncture monotherapy/acupuncture combined therapy), average duration of stroke (0–1 month/1–2 months/2–3 months), and treatment duration (less than 4 weeks/4 weeks or more). The results showed that none of these factors significantly contributed to the heterogeneity ($p > 0.05$) (see Table 2).

Sensitivity analysis using the one-by-one exclusion method was performed for the studies involving the PAS scale, and no study was found to have a strong impact on the results (see Figure 5).

Bias assessment was conducted for the studies involving the PAS scale, and the results showed a p-value of $0.519 > 0.05$, indicating no publication bias in this study (see Figure 6).

3.4.2 Overall effective rate

Nine studies (31–33, 36, 39, 40, 42, 44, 46) involved statistical analysis of total effective rate, totaling 612 cases included. Based on treatment duration, the analysis was divided into two subgroups: ≥4 weeks and <4 weeks. There was no statistically significant difference between the subgroups ($p = 0.25$, >0.05, $I^2 = 25.6\%$). The heterogeneity of the total effective rate was low ($I^2 = 0 < 50\%$, and the p value of the Q test was $0.92 > 0.1$), so a fixed-effects model was chosen for the meta-analysis.

For studies with treatment duration ≥4 weeks, the combined effect size was 4.71 (2.42, 9.15), and the effect size was significant ($Z = 4.56$, $p = 0.00 < 0.05$). For studies with treatment duration <4 weeks, the combined effect size was 2.47 (1.04, 5.86), and the effect size was significant ($Z = 2.05$, $p = 0.04 < 0.05$). The overall combined effect size was 3.77 (2.23, 6.36), and the effect size was significant ($Z = 4.97$, $p = 0.00 < 0.05$), indicating that the total effective rate in the intervention group was 3.77 times higher than that in the control group, demonstrating a significant intervention effect (see Figure 7).

Sensitivity analysis using the one-by-one exclusion method was performed for the studies involving the overall effective rate, and no study was found to have a strong impact on the results (see Figure 8).

Bias assessment was conducted for the studies involving the overall effective rate, and the results showed a p-value of $0.388 > 0.05$, indicating no publication bias in this study (see Figure 9).

3.4.3 Video fluoroscopic swallowing study

Four studies (36, 41, 42, 44) involved the statistical analysis of the VFSS scale, with a total of 306 cases included. The baseline consistency analysis showed low heterogeneity in the baseline effects of the VFSS scale between the experimental and control

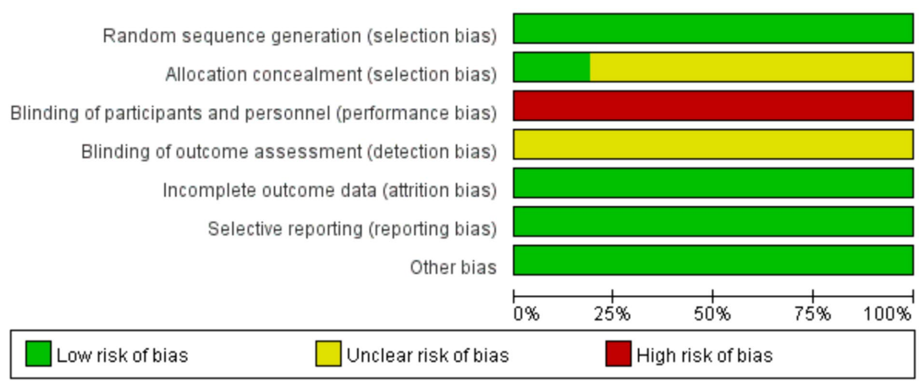

FIGURE 2
Risk of bias analysis.

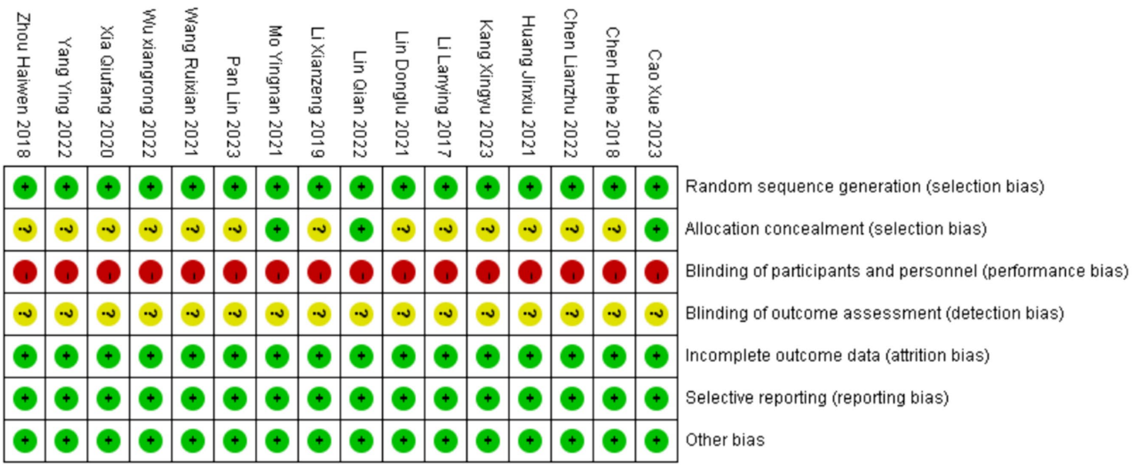

FIGURE 3
Summary of risk of bias.

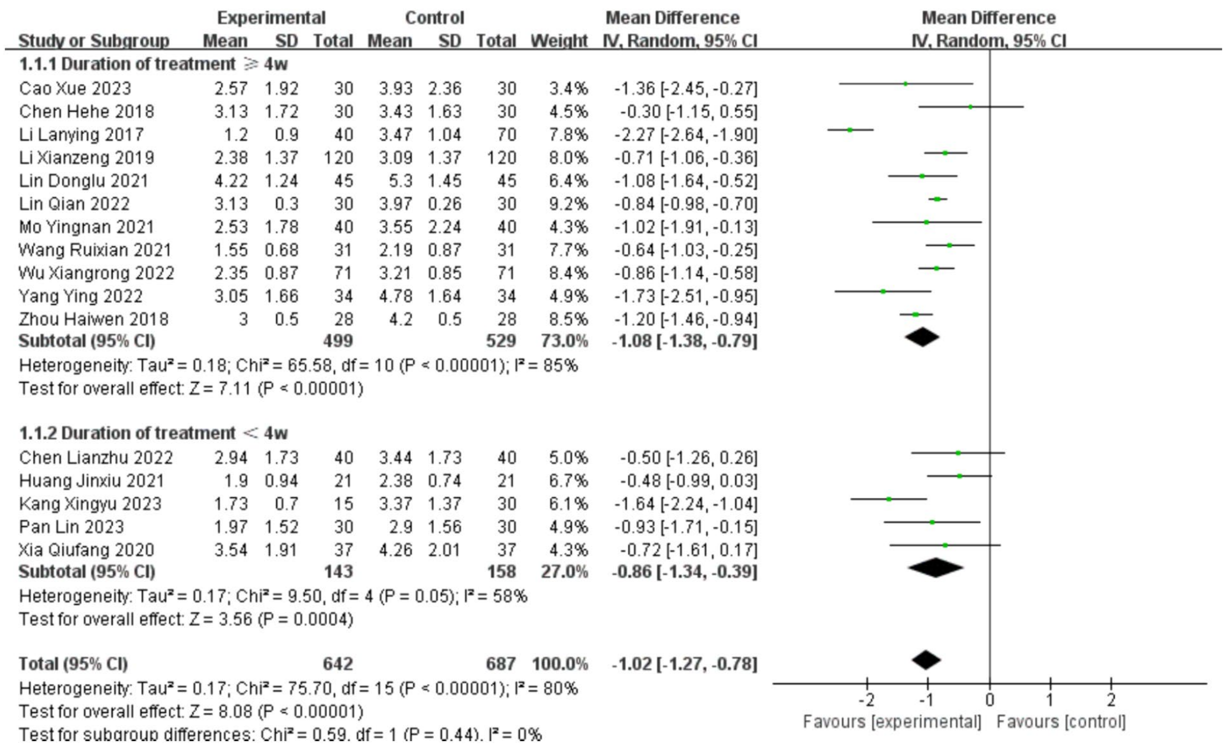

FIGURE 4
Forest plot of PAS scores.

groups ($I^2 = 0\% < 50\%$, and $p = 0.92 > 0.1$ from the Q test). Therefore, a fixed-effects model was used to combine the effect sizes. The combined effect size for the baseline effects was −0.08 ($Z = 0.67$, $p = 0.50 > 0.05$), indicating no statistically significant difference in the baseline period, allowing for subsequent meta-analysis (see Supplementary Figure S2).

According to the heterogeneity analysis, there was high heterogeneity in the effect sizes after treatment ($I^2 = 96\% > 50\%$, and $p = 0.000 < 0.05$ from the Q test). Therefore, a random-effects model was used for the meta-analysis. The combined effect size was 1.32 (0.08, 2.55), and the effect size was significant ($Z = 2.09$, $p = 0.04 < 0.05$), indicating that the VFSS scale scores in the intervention group were significantly higher than those in the control group by 1.32. The intervention had a significant effect (see Figure 10).

Sensitivity analysis using the one-by-one exclusion method was performed for the studies involving the VFSS scale, and no study was found to have a strong impact on the results (see Supplementary Figure S3).

TABLE 2 Meta-regression of PAS scores.

_it	Coef.	Std. Err.	*t*	*P* > |*t*|	[95% Conf.Interval]	
Year	−0.02647	0.091943	−0.29	0.779	−0.22883	0.1758981
Interventions	−0.12439	0.356066	−0.35	0.733	−0.90808	0.6593082
Course	0.099043	0.373485	0.27	0.796	−0.72299	0.9210771
Average course of illness	0.286838	0.201132	1.43	0.182	−0.15585	0.7295267
_cons	51.80965	185.7511	0.28	0.785	−357.026	460.6451

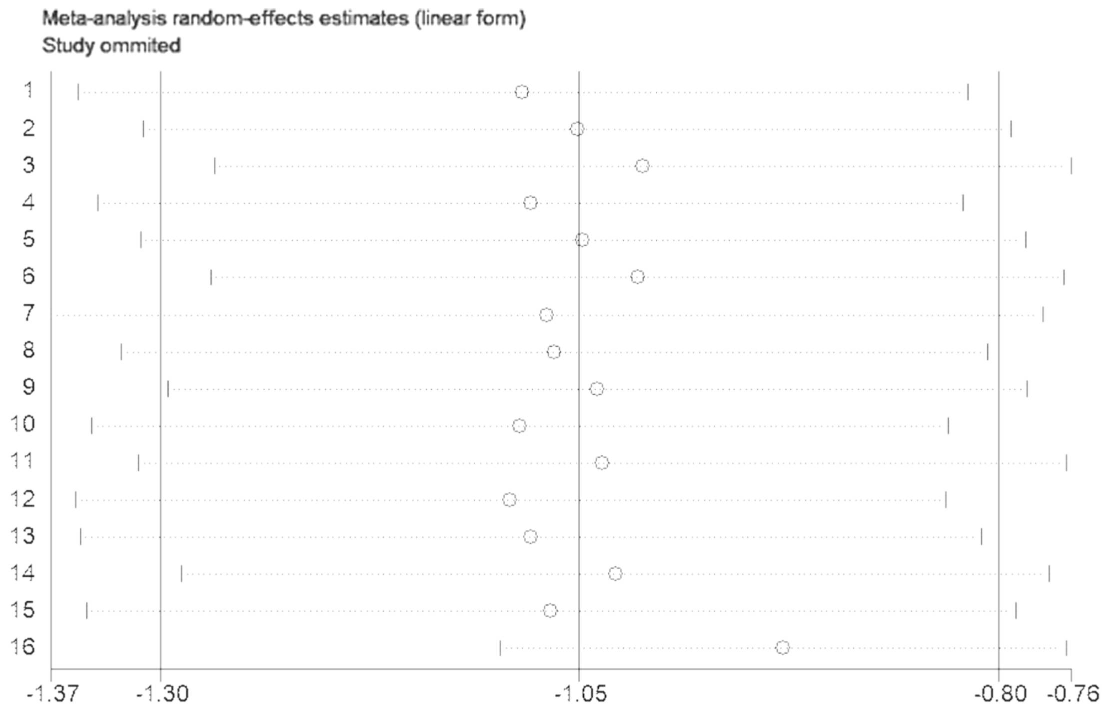

FIGURE 5
Sensitivity analysis of PAS scores.

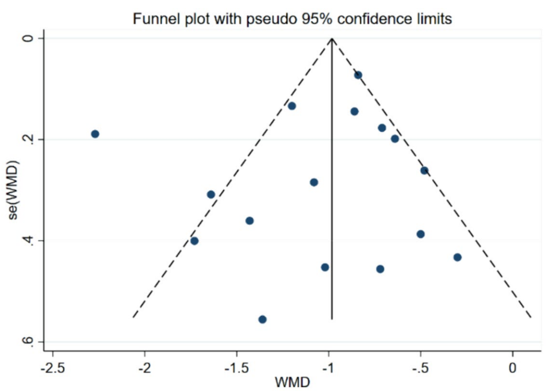

FIGURE 6
Funnel plot of PAS scores.

3.4.4 Hyoid bone displacement

Three studies (38, 40, 43) involved the statistical analysis of hyoid bone displacement, with a total of 312 cases included. The studies were divided into two subgroups based on the distance of hyoid bone upward displacement and forward displacement. The baseline consistency analysis showed low heterogeneity in the baseline effects of hyoid bone displacement between the experimental and control groups ($I^2 = 0\% < 50\%$, and $p = 0.89 > 0.1$ from the *Q* test). Therefore, a fixed-effects model was used to combine the effect sizes. The combined effect size for the baseline effects was 0.02 ($Z = 0.12$, $p = 0.90 > 0.05$), indicating no statistically significant difference in the baseline period, allowing for subsequent meta-analysis (see Supplementary Figure S4).

According to the heterogeneity analysis, there was low heterogeneity in the effect sizes after treatment in the forward displacement group ($I^2 = 0\% < 50\%$, and $p = 0.38 > 0.05$), while there was high heterogeneity in the effect sizes after treatment in the upward displacement group ($I^2 = 97\% > 50\%$, and $p = 0.00 < 0.05$). The overall result showed high heterogeneity ($I^2 = 93\% > 50\%$, and $p = 0.00 < 0.05$). Therefore, a random-effects model was used for the meta-analysis. The combined effect size was 2.02 (0.86, 3.18), and the effect size was significant ($Z = 3.41$, $p = 0.00 < 0.05$), indicating that the hyoid bone displacement in the intervention group was significantly higher than that in the control group by 2.02 mm. The intervention had a significant effect (see Figure 11).

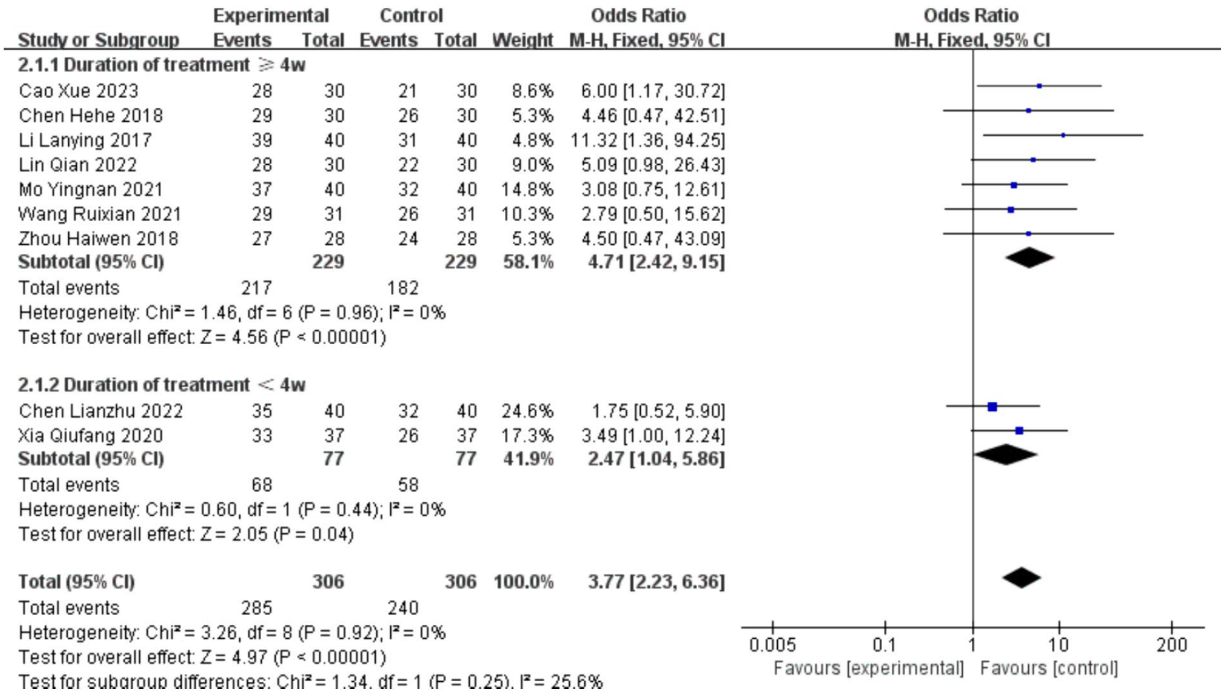

FIGURE 7
Forest plot of overall effective rate.

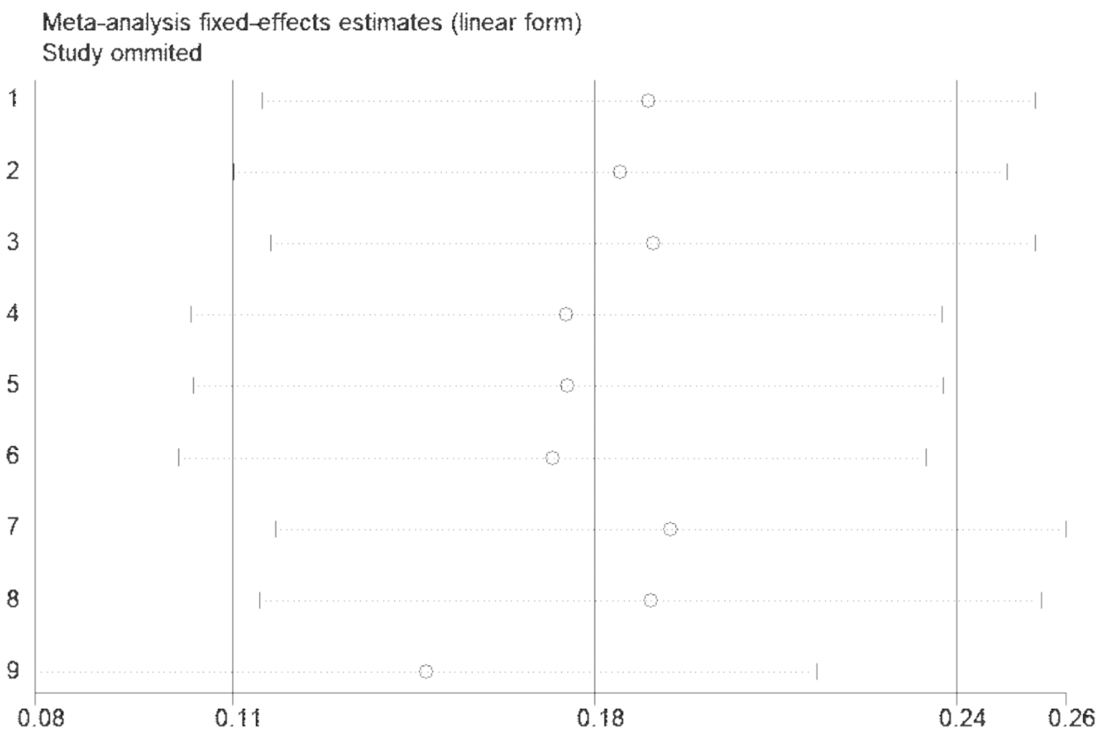

FIGURE 8
Sensitivity analysis of overall effective rate.

Sensitivity analysis using the one-by-one exclusion method was performed for the studies involving hyoid bone displacement, and one study (40) was found to have a strong impact on the results (see Supplementary Figure S5).

3.5 Safety analysis

Four studies (39, 40, 42, 45) reported adverse events or safety evaluations related to acupuncture. One study (42) reported one

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case of subcutaneous hematoma and one case of mild nasal mucosal bleeding, both of which were resolved in the short term through timely treatment. The safety evaluations in three studies (39, 40, 45) did not report any adverse events.

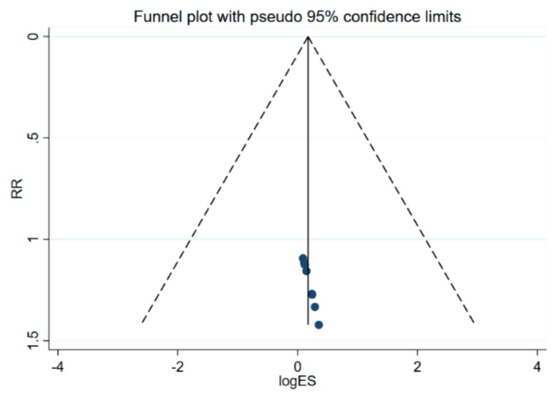

FIGURE 9
Funnel plot of overall effective rate.

4 Discussion

4.1 Summary of main results

This study included a total of 16 studies. The results found that compared with swallowing training, mirror therapy, urinary catheter balloon dilation, rTMS, and NMES, acupuncture alone or in combination therapy appears to be more effective in improving aspiration caused by PSD. However, caution is warranted in interpreting the results of this study. Firstly, among the included studies, 13 studies (32–38, 41–46) were identified as high risk of bias, and 3 studies (31, 39, 40) were categorized as unknown risk of bias. Secondly, in this meta-analysis, many potential variables were not disclosed, which could have varying degrees of impact on the results. This resulted in significant heterogeneity in the study. Nevertheless, it is noteworthy that the heterogeneity analysis of the primary outcome indicators in this study showed statistically significant differences, indicating the true efficacy of acupuncture for aspiration caused by PSD.

There was considerable heterogeneity in the primary outcome indicators in the studies. Subgroup analysis was attempted based on treatment duration, but the results still exhibited high heterogeneity. Subsequently, meta-regression was conducted based on publication year, intervention measures, average disease duration, and treatment course,

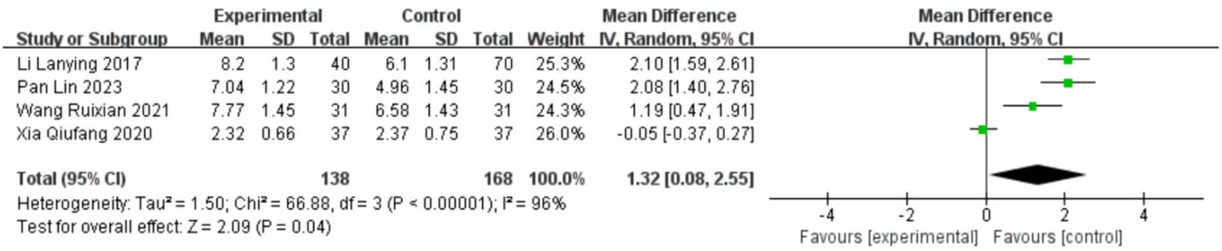

FIGURE 10
Forest plot of the VFSS scale.

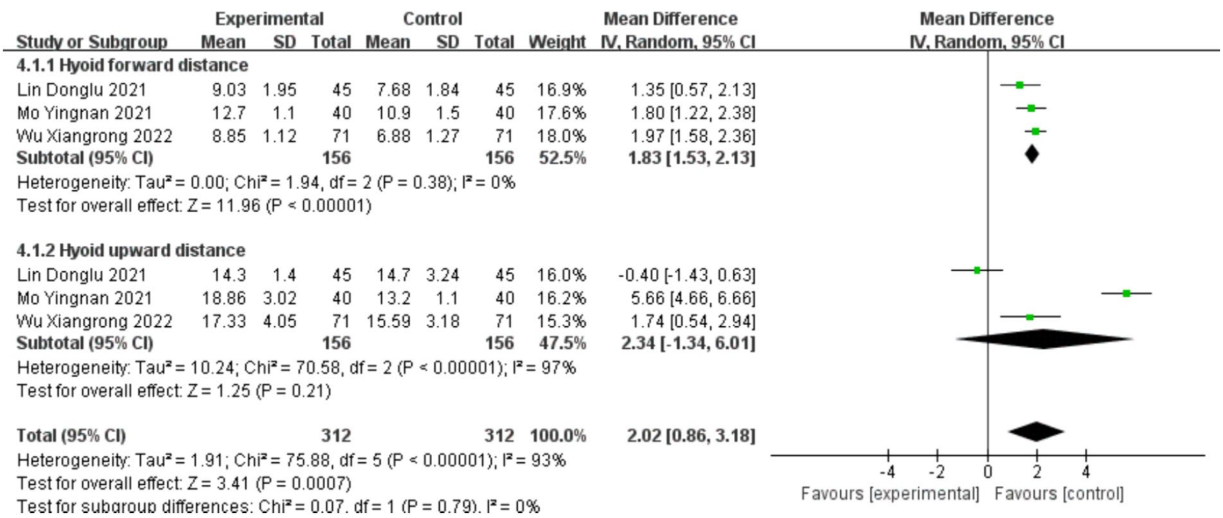

FIGURE 11
Forest plot of hyoid bone displacement.

revealing that these factors were not the cause of the high heterogeneity. Therefore, the origin of heterogeneity may be related to the type and location of stroke, as different types and locations of strokes have varying impacts on swallowing function, directly affecting the efficacy of acupuncture treatment and indirectly influencing the results of this study. Unfortunately, despite most studies reporting the number of cases of ischemic and hemorrhagic strokes, they did not separate the data when reporting results, making it impossible to verify this hypothesis. Additionally, the characteristics of acupuncture therapy itself may also contribute to the heterogeneity. The efficacy of acupuncture is influenced by factors such as acupoint selection, needle insertion time, and manipulation techniques, which are challenging to standardize across studies. Moreover, the swallowing training used in control groups in some studies is influenced by factors such as physician skill level and patient compliance, while the instrument brand and treatment intensity used in studies involving rTMS and NMES also greatly impact the study results.

Sensitivity analysis showed that apart from one study (40) having a significant impact on the analysis of hyoid bone mobility, the rest of the studies did not affect the results, possibly due to the small number of included studies. Adverse reactions to acupuncture mainly included subcutaneous hemorrhage or hematoma, without serious side effects. However, only 4 studies (39, 40, 42, 45) involved safety analysis of acupuncture treatment for post-stroke aspiration, indicating insufficient evidence regarding the safety of acupuncture treatment for aspiration caused by PSD.

In conclusion, acupuncture can be considered as a treatment method for aspiration caused by PSD. However, due to the high risk of bias and heterogeneity in the included studies, the efficacy of acupuncture treatment for aspiration caused by PSD still requires further validation.

4.2 Comparison with other systematic reviews

Different conclusions were found in other similar systematic reviews. A Cochrane systematic review published in 2008 (47) investigated the efficacy of acupuncture in treating acute post-stroke dysphagia and found insufficient evidence to draw any conclusions regarding the efficacy of acupuncture. However, this review only included one study with a total of 66 participants, which had a small sample size and outdated research results. More updated studies are needed to provide valuable evidence in evidence-based medicine. Another Cochrane systematic review published in 2018 (48) evaluated the efficacy of eight different treatment methods, including acupuncture. The results showed that swallowing treatment did not reduce PAS scores. However, this review did not have subgroup effects, and therefore, the results represented a general analysis of the overall efficacy of multiple treatment methods and cannot be used as independent evidence for the efficacy of acupuncture. A systematic review published in China in 2020 (49) demonstrated significant efficacy of acupuncture single therapy and acupuncture combined therapy, similar to the results of this study. However, the chosen outcome measures in that study did not include quantitative evaluation indicators for aspiration, and therefore, the research results cannot serve as evidence for the efficacy of acupuncture in treating aspiration caused by post-stroke dysphagia. Considering the results of previous systematic reviews, the efficacy of acupuncture in treating post-stroke aspiration remains controversial. The Chinese systematic review aligns with the results of this study. This may be related to the study period or the ease of including clinical randomized controlled trials conducted in China in the Chinese systematic review, as acupuncture is more widely used in China, and clinicians may have more experience and patients may have higher compliance, among other factors.

4.3 Mechanism studies

Normal swallowing activity consists of the oral preparatory phase, oral phase, pharyngeal phase, and esophageal phase, with the pharyngeal phase being the most critical and the stage where aspiration is most likely to occur. The key determinant of the pharyngeal phase of swallowing is the movement of the hyolaryngeal complex, which plays a crucial role in preventing food bolus entry into the airway and facilitating its entry into the esophagus (50). During the swallowing process in the pharyngeal phase, the hyoid bone and larynx undergo elevation and anterior movement. The anterior movement opens the upper esophageal sphincter, while the elevation causes epiglottic retroversion and closure of the larynx, allowing the food bolus to pass through (51). Therefore, analyzing hyoid bone displacement can provide insights into the completion of swallowing actions and the likelihood of aspiration.

Swallowing disorders result from damage to the swallowing motor area and its connection with the brainstem, leading to difficulties or inability to swallow (52–54). Although the swallowing reflex relies on the brainstem swallowing center, the initiation of swallowing is an autonomous activity (55). The swallowing neuronal network in the brainstem receives signals from the medullary swallowing center, and various cortical regions, including the insula, primary motor cortex, and somatosensory cortex, play important regulatory roles in swallowing (53). PSD is mainly caused by lesions in the swallowing cortical center, subcortical fibers, medullary swallowing center, and extrapyramidal system (2).

Acupuncture can enhance the excitability of the central nervous system, coordinate fine movements of the tongue and pharynx, and relieve pharyngeal muscle paralysis, thereby improving PSD (56). Research by Fangfang Fang and colleagues has shown that acupuncture may regulate the cortical swallowing area to the periaqueductal gray matter pathway by increasing the expression of c-Fos + neurons in the intact-side cortical swallowing area, calcium activity of neurons in the intact-side cortical swallowing area, and activity of neurons in the nucleus ambiguus, thereby achieving the therapeutic effect on PSD (57). Research by Junheng Shi has shown that in rats receiving acupuncture, a higher expression of c-Fos protein was observed in the motor neurons of the nucleus ambiguus, indicating that acupuncture can transmit impulses to the swallowing motor neurons of the nucleus ambiguus, activating the neurons and expressing immediate-early response proteins such as c-Fos. This may also be one of the central nervous system mechanisms of acupuncture in treating PSD (58).

4.4 Advantages and limitations

Advantages of this study: The research process strictly followed the requirements for systematic reviews and meta-analysis outlined in the Cochrane Handbook for Systematic Reviews of Interventions 6.4 (59). The study included the latest clinical randomized controlled trials, and the outcome measures relied on direct judgments from imaging examinations, avoiding subjective evaluation errors from bedside scales.

Limitations of this study: In the search strategy, to ensure comprehensive retrieval of studies, the search strategy did not include "aspiration" as a subject heading or free term. This was because it was found during the search that most studies related to acupuncture treatment for post-stroke dysphagia did not mention terms related to "aspiration" in the title or abstract. Therefore, to minimize the risk of missing studies, some sacrifices were made in the accuracy of the search strategy in this study. The high heterogeneity among the included studies made it difficult to identify the source through subgroup analysis and meta-regression. The heterogeneity in this study arose during the establishment of inclusion and exclusion criteria, which is related to the characteristics of acupuncture therapy itself. Most of the included studies lacked safety assessment and recording of adverse events, and did not provide explanations for sample size calculation methods and processes. Additionally, all the literature included in this study was published in Chinese, so the conclusions drawn need to be interpreted cautiously, as they may only indicate the efficacy of acupuncture combined therapy and acupuncture single therapy for treating aspiration caused by post-stroke dysphagia in China.

4.5 Implications for future research

Future research should provide more detailed reporting of experimental design, safety evaluations, sample size calculations, and other aspects to improve research quality. Case selection can be further refined, such as including only patients with a specific type of stroke to enhance the study's specificity. Higher-quality efficacy evaluation indicators with stronger evidence should be chosen to minimize errors resulting from indirect and subjective evaluations. More clinical randomized controlled trials on post-stroke aspiration, especially for silent aspiration, should be conducted to provide a foundation for higher-quality evidence in evidence-based medicine.

5 Conclusion

Both acupuncture combined therapy and acupuncture single therapy can effectively improve aspiration caused by post-stroke dysphagia, with a low incidence of adverse events. However, due to the low quality of the included literature, more high-quality randomized controlled trials are still needed to confirm the effectiveness and safety of acupuncture in the treatment of aspiration caused by post-stroke dysphagia.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

HL: Data curation, Formal analysis, Investigation, Software, Visualization, Writing – original draft, Writing – review & editing. JL: Conceptualization, Data curation, Resources, Supervision, Writing – review & editing. XW: Conceptualization, Methodology, Supervision, Writing – review & editing. ZZ: Conceptualization, Formal analysis, Funding acquisition, Investigation, Supervision, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fneur.2024.1305056/full#supplementary-material

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Mapping research trends regarding the mechanism of dysphagia from 1993 to 2023: a bibliometrics study and visualization analysis

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As a common consequence of various neurogenic disorders, dysphagia has a significant impact on the quality of life for patients. To promote the development the field of swallowing, it will be helpful to clarify the pathological and therapeutic mechanisms of dysphagia. Through visual analysis of related papers from 1993 to 2023 in the Web of Science Core Collection (WoSCC) database, the research status and development trend of the pathogenesis of dysphagia were discussed. The co-occurrence study was finished using CiteSpace 6.2 R4 software, including keywords, countries, institutions, and authors. Finally, 1,184 studies satisfied the inclusion requirements. The findings of the visualization analysis suggested that aspiration and gastroesophageal reflux disease would be the areas of greatest interest for researchers studying the mechanism of dysphagia. As for the latest occurred research trends, fMRI, signals and machine learning emerging into the field of view of researchers. Based on an analysis of country co-occurrence, United States, Japan and China rank the top three, in terms of the number of publications on dysphagia. University System of Ohio is the organization that has published the most amount of articles regarding the mechanism of dysphagia. Other highly published schools in the top three include State University System of Florida and Northwestern University. For the prolific authors, German, Rebecca Z published the most articles at present, whose own research team working closely together. Several closely cooperating research teams have been formed at present, including the teams centered around German, Rebecca Z, Warnecke, Tobias and Hamdy Shaheen. This study intuitively analyzed the current research status of the mechanism of dysphagia, provided researchers with research hotspots in this field.

KEYWORDS

dysphagia, mechanism, CiteSpace, visualization analysis, research frontiers, emerging trends

Introduction

Dysphagia is a clinical manifestation in which the structure or function of the mandible, lips, tongue, soft palate, throat, esophagus and other organs are impaired, leading to the food cannot be safely and effectively transported from the mouth to the stomach (1). Inability to eat, delayed swallowing, or aspiration of food are characteristics of dysphagia. Dysphagia occurs in up to 81% of patients after stroke or 80% of Parkinson's disease (PD) (2, 3), and can also occur in other neurogenic diseases (4–6), such as head and neck tumor surgery (7). According to research, dysphagia also occurs in the normal people over the age of 60, with a prevalence of about 40% (8).

However, the incidence of dysphagia is influenced by the means of assessment and may be greater depending on the method, timing and criteria used to diagnose dysphagia. In the acute phase, gold-standard routine tests such as VFFS and FEES have practical limitations in that they can only detect obvious dysphagia and therefore miss silent inhalations (9). Therefore, various of measurement have been carried out to correctly diagnose dysphagia, such as bedside evaluation, which can improve the diagnosis of aspiration in the elderly, with high sensitivity in acute stroke patients (10, 11). At the same time, dysphagia after stroke is also affected by a variety of factors, such as NIHSS value, cognitive dysfunction, and the degree of white matter disease, which were described as independent predictors of post-stroke dysphagia and persistent dysphagia at 14 days. So was the pneumonia post-stroke (12, 13).

Dysphagia can lead to a variety of adverse complications, for instance, aspiration, malnutrition, dehydration, and asphyxia (14). It can even lead to serious consequences, such as aspiration pneumonia or death from choking on large food pellets. For those complications, aspiration is easily overlooked, yet many studies have focused on dysphagia without considering aspiration (15). Therefore, highly sensitive dysphagia bedside screening tests (BSEs) designed to detect aspiration and tested against FEES are more likely to describe the real incidence rate (13, 16–18). However, most dysphagia screening often fail to detect silent aspiration, which can cause most dysphagia associated pneumonia (13). Specifically, diagnosing dysphagia after stroke by focusing on obvious signs of aspiration, such as coughing or voice changes, can lead to silent aspiration going undiagnosed, increasing the relative risk of pneumonia and resulting in a poor clinical prognosis due to false negative results (13). Therefore, more clinical attention to screening aspiration, especially silent aspiration, are particularly important. Actually, the combination of BSEs and other methods can improve the detection rate and avoid the misdiagnosis of silent aspiration. As far as we know, there has been research on the detection of silent aspiration, and the project is currently in progress (19). Due to the complexity of its etiology and serious consequences, dysphagia attracted the attention of researchers, and more and more studies on the mechanism of dysphagia are emerging.

In recent years, studies focused on the mechanism of dysphagia have been conducted from the perspectives of central and peripheral. With the in-depth research and increasing interest in the field of swallowing disorders, studies focused on the current situation and trends in this field are needed. After reviewing the literature, we found four related visualization studies that payed attention to the dysphagia (20), post-stoke dysphagia (PSD) (21, 22) or rehabilitation of PSD (23). Those studies analyzed the current literature or the rehabilitation on dysphagia or PSD to identify the research hotspots and frontiers using bibliometrics. However, until now, scientometric analysis in the mechanism of dysphagia has rarely been reported. CiteSpace is one of the most widely used visualization tools, as a large amount of data have been transformed into more intuitive visual maps that can reveal the frontiers and trends in certain fields. It is an influential software in the field of information mapping and visualization that can display the knowledge domain of related disciplines within a certain period of time through intuitive visualization analysis (24). CiteSpace intuitively highlights the quantitative features of research articles in the field, helping researchers understand the evolution of certain field over time (25).

In our study, we used CiteSpace software to uncover cooperative networks and evaluate the research frontiers and emerging trends in the mechanism of dysphagia, and a visual map of keywords, countries, institutions, and authors was generated. Therefore, we will comprehensively and objectively integrate and elaborate the core and frontier information in the field of dysphagia to provide objective and visualization data for relevant researchers.

Methods

Data sources

Studies retrieval: the search terms for the topics of study were "dysphagia" or "swallowing dysfunction" or "swallowing disorders" and "mechanism", which were searched from the Web of Science Core Collection (WoSCC) database. The search period was from January 1, 1993, to November 30, 2023. Selection: studies on humans or animals of mechanism on dysphagia were both included. The research type is defined as original research. Exclusion: review articles, editorial material, meeting abstract, early access, note, book chapters, letter, retracted publication and correction. The research process was as follows (Figure 1): a total of 1,609 relevant studies were initially retrieved, 341 documents that did not fit the type were excluded. And 84 irrelevant documents were excluded through examination and abstract reading. As a result, 1,184 papers were finally included.

Data processing

The raw data from WoSCC were searched and downloaded, and the title and abstract were screened by two independent reviewers (Qiuping Ye and Jiahui Hu). The articles included in the study were exported from the WoSCC database in plain text format and named separately in the download_xxx.txt way. The exported information of the articles included title, year of publication, author, research institution, keywords, abstract, journal, etc. CiteSpace 6.2. R4 software (Drexel University, Philadelphia, PA, USA) was used to process and transform the data.

Statistical methods

All data were analyzed in CiteSpace 6.2. R4 software, and the node type included the author, institution, and time span. The time span of the keyword node was set to 3 year. The selection criteria were set to the g index $k = 25$, the top 50% level of the most cited or occurred items from each slice. Statistical analysis of keyword co-occurrence analysis, keyword clustering, country

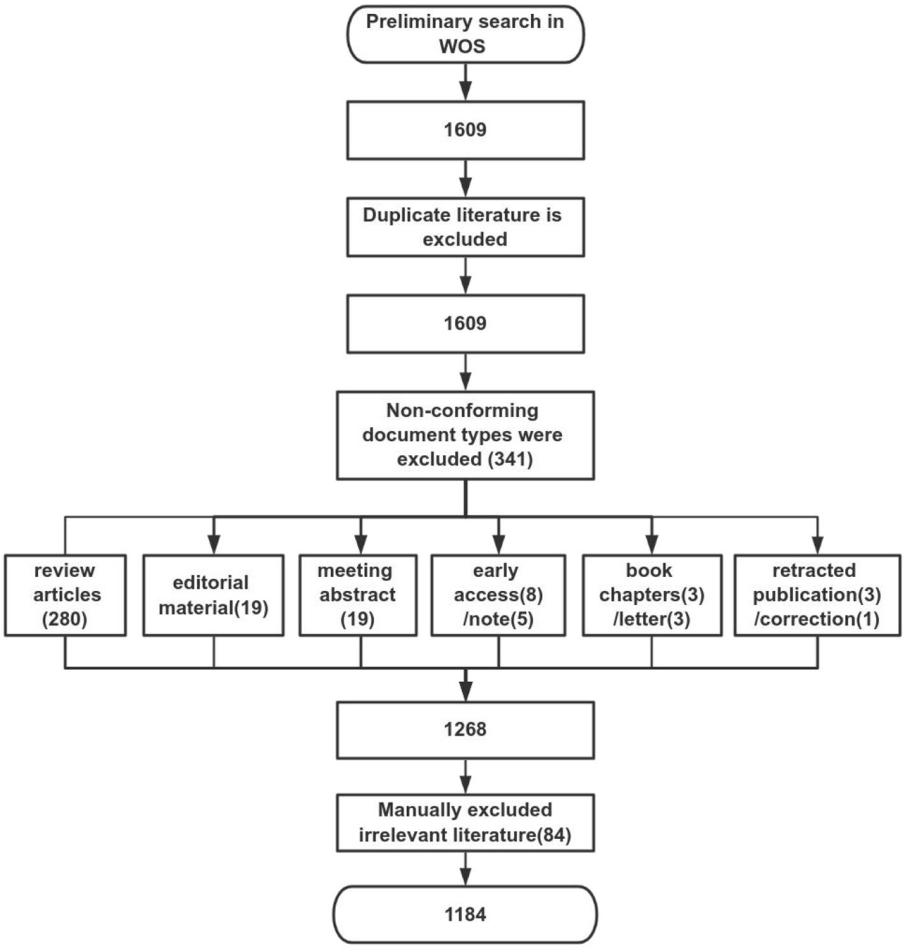

FIGURE 1
The study tree of the whole research.

and institution co-occurrence analysis, and author co-occurrence analysis, were all processed by CiteSpace. The concrete principles of these methods refer to previous studies (26), which indicated three types of analysis: network visualization, overlay visualization, and density visualization. The nodes in each knowledge graph represented different publications, and the size of the nodes was proportional to their frequency of occurrence in a specific period. Similarly, the connection between nodes indicates the degree of relationship; the thicker the lines are, the stronger the connections. These analyses will make the study field more intuitive, allowing people to observe the trends of various research hotspots over time (27).

Results

To some certain extent, the development level and research popularity of the field can be inferred from the statistics on the number of published papers each year. From 1993 to 2023, a total of 1,184 literature were published. This field saw modest growth from 1994 to 2022 and a downward trend in 2023 as a result of incompletely obtained data from 2023. Up until the point of the literature search, the number of publications reached its peak in 2022 with 89 citations. Figure 2 depicted a stable rise during the entire development phase.

Co-occurrence analysis of keywords

Keywords are a high summary of the core content of the article, and the research hotspots in this field can be judged by the co-occurrence analysis of keywords. In this study, the top 50 most-cited or -occurring items were chosen from each slice. Following the co-occurrence of keywords, 465 nodes and 746 lines were ultimately produced; more than 14 keywords appeared more than 50 times, with the top 10 appearing in Table 1. As illustrated in Figure 3, nodes stand in for keywords, and each node's size reflects how frequently the keywords occur together. There was a strong correlation between the keywords in this article, as evidenced by the line that shows the strength of the link between them. When paired with Table 1, the highest frequency was 108 for "gastroesophageal reflux disease," 100 for "aspiration," and "dysfunction," with

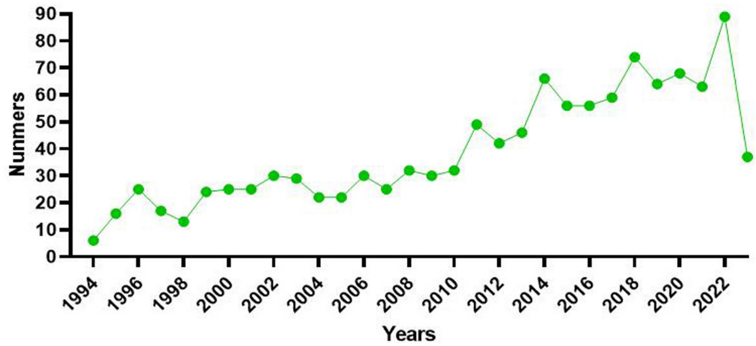

FIGURE 2
The number of publications from 1993 to 2023 is described by a line chart. The green points indicated the numbers of publications.

TABLE 1 The top 10 keywords in the co-occurrence analysis.

Rank	Count	Centrality	Keywords
1	362	0.04	Dysphagia
2	249	0.05	Mechanism
3	108	0.07	Gastroesophageal reflux disease
4	100	0.05	Aspiration
5	100	0.12	Dysfunction
6	77	0.08	Management
7	67	0.11	Oropharyngeal dysphagia
8	63	0.15	Children
9	60	0.05	Stroke
10	52	0.02	Parkinson's disease

the exception of dysphagia (362) and mechanism (249). As for the latest keywords, fMRI, signals and machine learning occurred more frequent (Table 2), which indicating more new research techniques and directions emerged in the mechanism of dysphagia. It is worth noting that, dysphagia caused by interior cervical discectomy is a new vision for researchers, although only once, but in reminding our researchers need to pay attention to dysphagia caused by various diseases. The colored nodes, which represented the keywords, showed the growing trends in the data; the larger the node, the more frequent it is, which indicated the emerging trends in the field of dysphagia. The results indicate a substantial correlation with other terms, with adults having the strongest betweeness centrality (0.23) among all the categories. Figure 4A displayed the top 25 strongest citation bursts. Citation bursts for gastroesophageal reflux disease was the strongest; they began in 1999 and ended in 2010. The red dots in Figure 4B that were obtained based on the results of Figure 3 also highlighted the recent emergence of penetration aspiration and post-stroke dysphagia in 2020. The historical progression of dysphagia research is depicted in Figure 4B, and the citation bursts in Figure 4A are indicated by the red nodes in Figure 4B.

Cluster analysis of keywords

Cluster analysis of keywords can reflect research hotspots and research frontiers in this field. We finally obtained 20 clustering tags, and the value of the keywords cluster module was (Q) = 0.773 > 0.3, indicating that this cluster structure was significant (Figure 5). The average silhouette was (S) = 0.9123 > 0.7, indicating that the members of the clusters had good homogeneity and high confidence. The first 10 cluster labels were retained in this study, including transcranial magnetic stimulation (TMS), achalasia, reflex, antireflux surgery, head and neck cancer, botulinum toxin, chemotherapy, eosinophilic esophagitis, substance p, and esophageal manometry. These groupings comprised molecular mechanisms, diseases, therapeutic strategies, and evaluation methodologies. Based on the included literature, we could determine that dysphagia caused by stomach and esophagus problems concerned the researchers. As for types of disease, researchers mainly focused on dysphagia after head and neck cancer except for stroke. Among those clusters, TMS occurred in a relatively new time in 2014, which indicated the research trends and hot spots for the treatment of dysphagia.

Distribution of countries and institutions

We were able to identify countries and institutions that have shown a greater interest in the mechanism of dysphagia and have made significant contributions to the field's progress through the examination of published literature. The co-occurrence analysis of countries and institutions reflects the distribution of major scientific research forces in this field. 61 nodes and 247 links were produced by the co-occurrence analysis atlas of the countries, of them, nine countries produced more than 50 articles (Table 3). The United States, Japan, and China are the top three contributors, accounting for 43% of all literature. Figure 6A depicted the international collaboration network, with link strength indicating the degree of cooperation between countries. USA was the most prolific country with 410 articles, followed by the Japan (116 papers) and China (92 articles). The most three contributing countries, however, have weak links and limited mutual cooperation, although they have all developed close

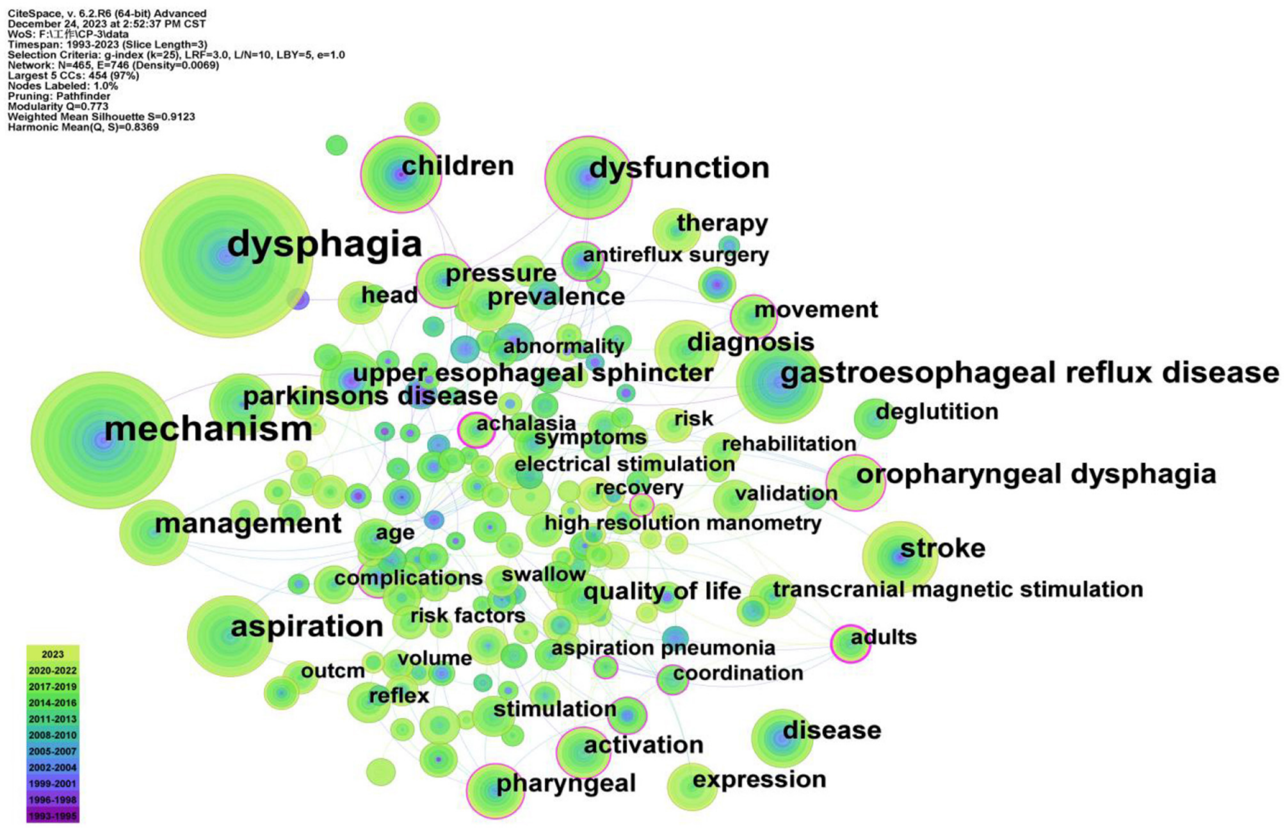

FIGURE 3
Co-occurrence analysis of the keywords. The thickness of the line indicates the strength of the connection between nodes. The node size indicates the frequency of keywords. In the figure, gastroesophageal reflux disease and aspiration occupied the main position except for dysphagia and mechanism.

TABLE 2 The latest keywords in the co-occurrence analysis.

Rank	Count	Centrality	Keywords
1	3	0.04	fMRI
2	2	0.03	Signals
3	2	0.02	Machine learning
4	2	0.02	MRI
5	1	0.02	Anterior cervical diskectomy

relationships with other countries. Therefore, in order to facilitate the sharing of beneficial resources and the advancement of the field, the major contributing nations should step up their cooperation.

With 339 institutions, the co-occurrence analysis atlas of institutions produced 339 nodes and 749 lines, showing a comparatively large number of research groups, the majority of which collaborated closely with each another. University System of Ohio, State University System of Florida, and Northwestern University were the top three significant output institutions, with respective outputs of 33, 31, and 31 (Figure 6B). However, 70% of the top 10 organizations came from the United States (Table 4), indicating an imbalanced distribution of research on the mechanism of dysphagia around the world. The three institutions with the most published papers are relatively independent in their research, which need more cooperation and exchange to promote the development of the industry.

Analysis of authors

The co-occurrence analysis of authors can reflect the cooperation and communication between academic leaders and authors in the field. In this study, the co-occurrence analysis of authors generated 411 nodes and 490 lines (Figure 7A). The top-ranked authors who have most publications were German, Rebecca Z with 14 documents (Table 5). The most citation documents of German, Rebecca Z was an article published in the journal of Dysphagia with 31 citation, which studied the relationship of unilateral superior laryngeal nerve (SLN) lesion and dysphagia and aspiration (28). However, his article published in the relative lower impact factor (IF) journal, among which 4 article published in the journal of Dysphagia (28–31) and 2 published in the journal of LARYNGOSCOPE (32, 33), both of which have the IF of 2.6. Dysphagia was an influential journal in swallowing and dysphagia field that has 5,546 total citations in 2022, while 28,923 total citations were seen in the journal of LARYNGOSCOPE. Authors who have second-ranked publications was Jadcherla, Sudarshan R

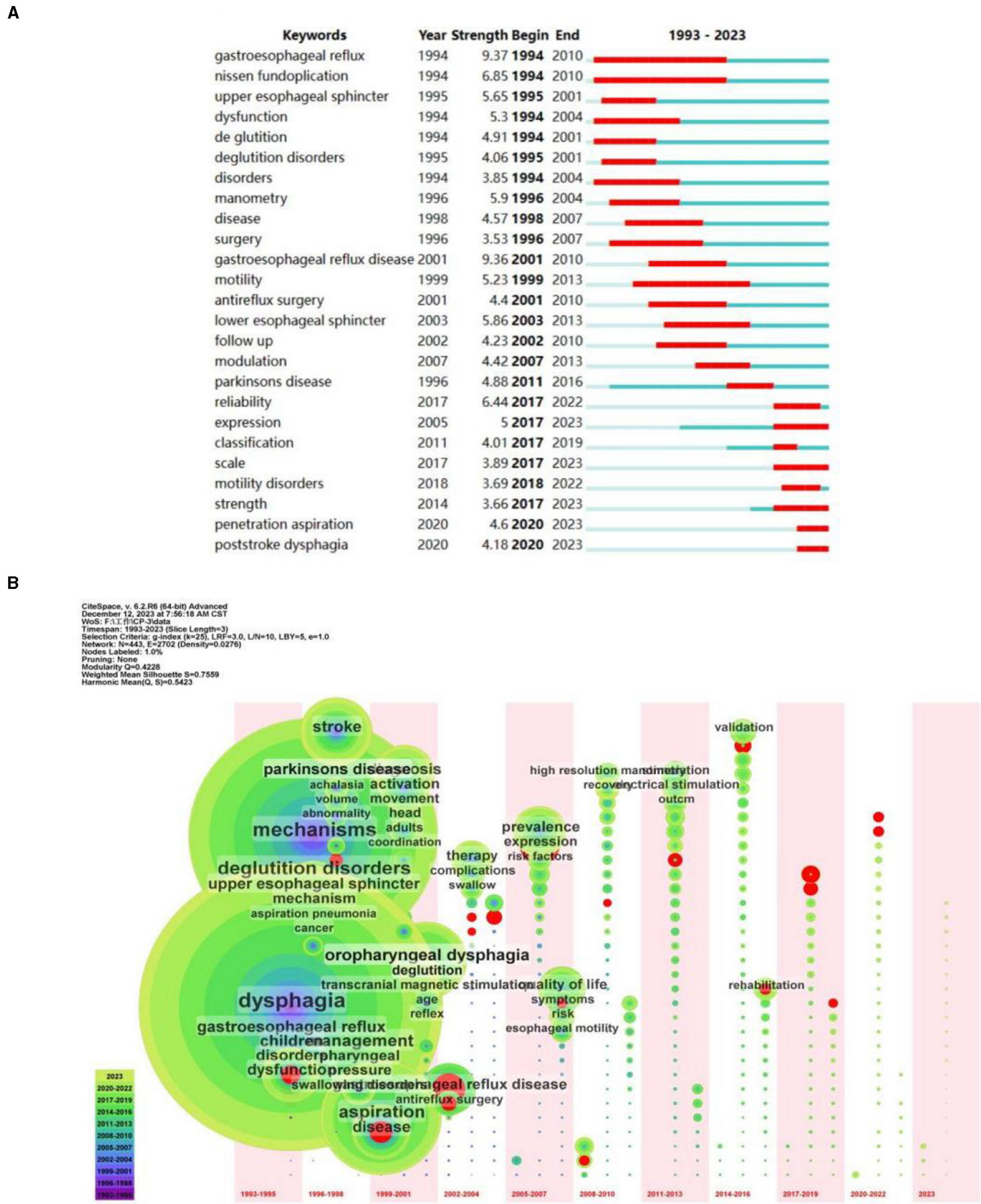

FIGURE 4
(A) The top 25 keywords with the strongest citation bursts. **(B)** The time zone of keyword clustering.

with 12 documents, following with Cock Charles, Steele Catriona M and Hamdy S, all of them has published 10 documents. On the top 10 authors who have most publications, Aydogdu I has most total citations with 625 and average citation with 69, following with Ertekin C with 334 total citations and 42 average citations. The highest IF journal they published is the journal of Brain with

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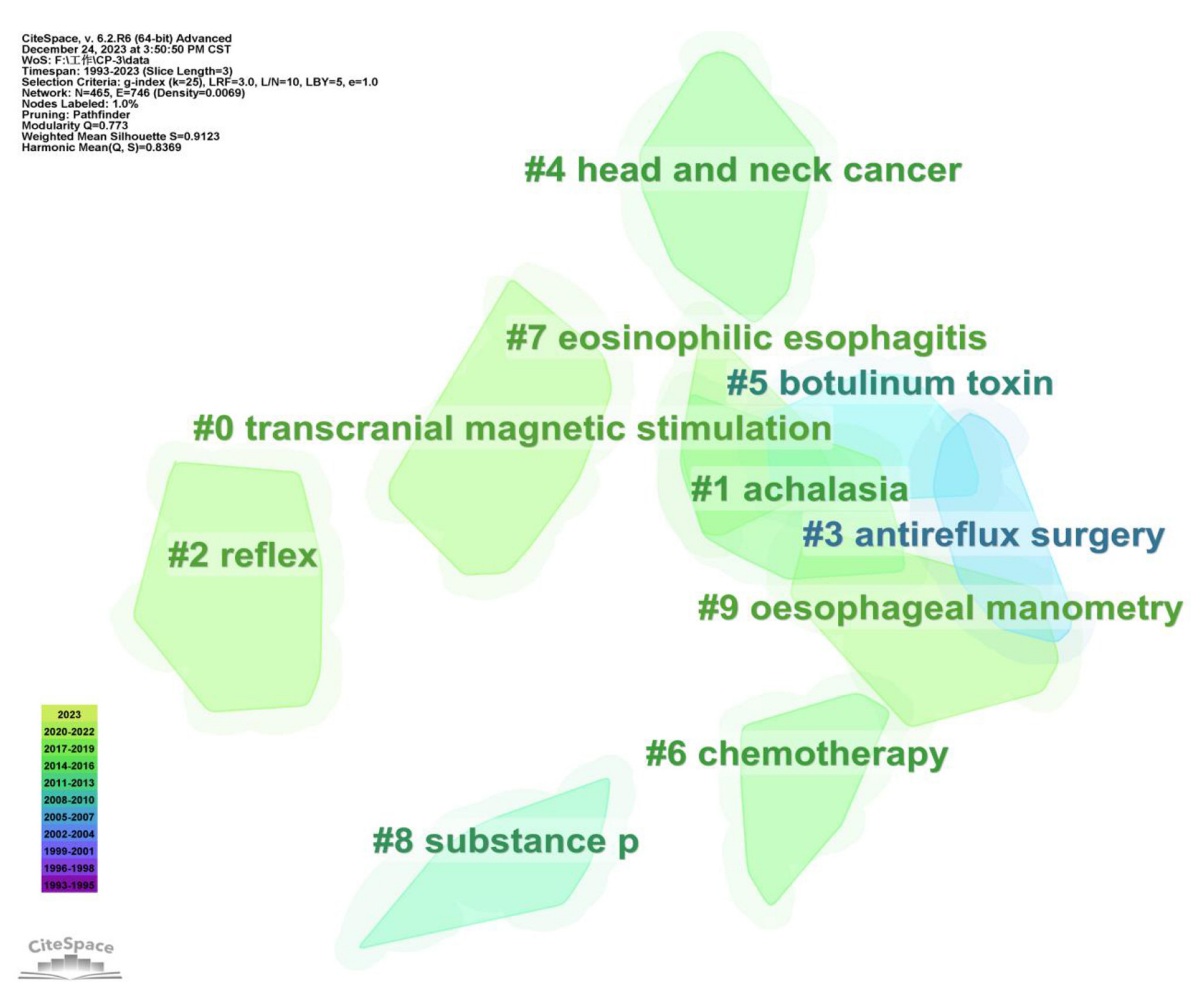

FIGURE 5
Cluster analysis of the keywords. In clustering, the same elements are aggregated together, each cluster exists independently, and the cluster label is a name for each cluster diagram. The top 10 larger cluster labels were retained in the figure. Different colors display different clusters. The largest three clusters are cluster 0, 1, and 2.

TABLE 3 The top 9 countries that published more than 50 articles in the co-occurrence analysis.

Rank	Count	Centrality	Countries
1	410	0.39	USA
2	116	0.02	Japan
3	92	0.02	People's Republic of China
4	79	0.34	England
5	70	0.07	Italy
6	66	0.24	Australia
7	66	0.12	Germany
8	54	0.1	France
9	54	0	South Korea

the IF of 14.5 in 2022, following with the journal of Neurology Neurosurgery and Psychiatry with the IF of 11.1. Furthermore, the average IF they published reached up to 6.82. Those data showed that they are the influential authors in this field. Interesting, they belong to the same team, which indicated that it is a team with close cooperation, rich results and high impact.

In additionally, there is little collaboration between the three most prolific authors, and they have their own independent research collaboration. Previous study showed that the top three authors with the most publications were Dziewas R, Hamdy S and Clave P (21), but two of them have published fewer than 10 research articles on the mechanisms of dysphagia. Similarly, the three teams with the largest volume of publications were decentralized and did not collaborate with each other. It is worth noting that the team centered around German, Rebecca Z and Gould, Francois D H formed a tight network model; Another tight collaboration team

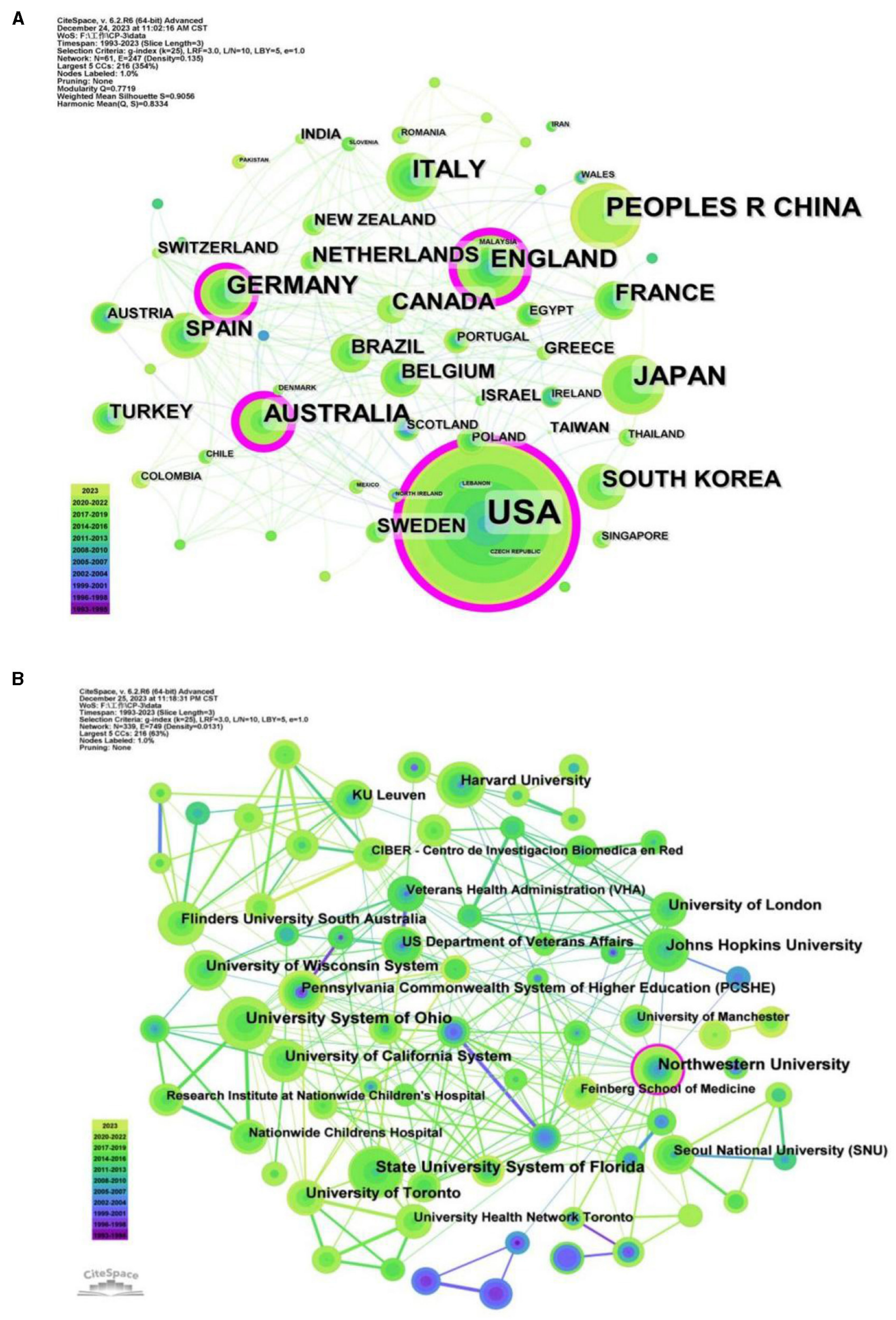

FIGURE 6
(A) Co-occurrence analysis of the countries. **(B)** Co-occurrence analysis of the institutions.

centered on Warnecke T and Dziewas R, Hamdy S and Michou E, Cock C and Omari, T (Figures 7B–D), among which, the team of Hamdy S and Michou E has high total and average citations. These figures reveal several stable groups of teamwork have been formed, including a collaborative research group centered on German Rebecca Z, Warnecke T, Hamdy S.

TABLE 4 The top 10 institutions in the co-occurrence analysis.

Rank	Counts	Centrality	Institutions
1	33	0.06	University System of Ohio
2	31	0.02	State University System of Florida
3	31	0.27	Northwestern University
4	25	0.04	Johns Hopkins University
5	24	0.05	University of California System
6	24	0.02	University of Wisconsin System
7	22	0.01	University of Toronto
8	22	0.07	University of London
9	20	0.07	Harvard University
10	19	0.03	Flinders University South Australia

Discussions

Dysphagia is a prevalent complication among neurogenic diseases, which seriously affect both the life of patients and the burden of family and society. Deeply analyzing the pathogenesis of diseases can help people take more effective preventive measures, which need mechanism researches. In this paper, relevant literature on the mechanism of dysphagia in WoSCC database in recent 30 years was used as the research object. They were visually analyzed by extracting the information about keywords, authors, countries, institutions. In the form of knowledge graph, we analyzed the current research status about mechanism of dysphagia from different perspectives, and discussed the research frontiers and trends in this field.

In the historical process of the mechanism of dysphagia, the study of central mechanism is the earliest and most studied, especially in stroke (34). We could also obtain that among diseases, stroke and Parkinson's disease appeared in the top 10 co-occurrence keywords, and they also have relative strongest citation bursts (Figure 4A, Table 1). For dysphagia after stroke, mechanism focused mostly on swallowing primary motor cortex (35) and excitatory neurons in swallowing related brains, including cortex and subcortical brain areas (34, 36, 37). Recently, the cerebellum has been considered as another regulatory brain region in recovery of dysphagia and may play an important role in bidirectional regulation (38). It is worth noting that the cortical compensation mechanism, such as the pharyngeal representations of unaffected hemisphere are important, which have been proposed in Gastroenterology in 1998 (39). This direction is still being investigated in the treatment of dysphagia until now. With the development of science and technology, neural circuits have attracted the attention of researchers (40), so various neuroscience methods, including optogenetics, chemogenetics and two-photon imaging, have been applied in this process.

For the latest keywords in the co-occurrence analysis of dysphagia, we could see that some emerging vision come into our view, including fMRI, signals and machine learning (Table 2). In the signal transmission of swallowing brains, excitatory neurotransmitters have received the most attention (34), while gamma aminobutyric acids (GABAs) is another neurotransmitter that can inhibit the activation of swallowing (41) and it also involved in suppressing the swallowing reflex after harmful irritation of the facial and oral structures (42). However, there has been little research in recent years. Furthermore, increases in exercise-related BDNF and tyrosine kinase receptor B (TrkB) may play a role in the mechanism that promotes increased tongue strength in young and middle-aged rats (43). In a previous study, 5-HT in the NTS also played a critical role in EA treatment of swallowing (44). In PD, the recovery of dysphagia may be associated with reduced presynaptic dopaminergic integrity in the caudate nucleus (45), but other researchers have not endorsed this idea (46). In recent years, gene regulation has attracted the attention of researchers (47), which needs in-depth study. The researchers proposed that the cerebral intestinal axis (48) and genes, such as polymorphisms of the brain-derived neurotrophic factor (BDNF) gene (49, 50), are associated with the recovery of dysphagia. In general, there are more and more studies on the mechanisms of swallowing, providing more scientific basis for the revealing the mechanism of dysphagia.

The results of cluster analysis of the keywords showed that TMS occupied an important category. As a non-invasive brain stimulation for the rehabilitation of dysphagia, TMS, especially repetitive TMS (rTMS) was mostly used. The group that has studied rTMS the most is Hamdy S 'team, whose research focuses mostly on the human pharyngeal motor cortex (51, 52). Those studies uncovered the underlying mechanism of rTMS treatment referring to the excitatory of cortex and cortical plasticity. Furthermore, except for the cortex, rTMS also regulates the cerebellum, which also increased the excitatory of pharyngeal motor cortex (53). However, the same stimulation on cerebellar vermis with rTMS took an opposite inhibitory effect to pharyngeal motor cortical activity and swallowing behavior (54). The effect of cerebellar on dysphagia attracted the attention of more and more researchers in recent year, which is a research frontier of the underlying mechanism of dysphagia. Researchers thought that TMS is a neurostimulatory techniques hold future therapeutic promise.

Limitations

Our study had some limitations. First, we only concentrated on visualization analysis of the mechanism of dysphagia, but did not distinguish and mention the mechanisms of specific treatment methods for dysphagia. Second, since the database we searched was only WoSCC database, the literature included was not comprehensive. More in-depth and comprehensive studies should be carried out in the future.

Conclusions

Research on aspiration concerned the researchers and practitioners, indicating the important of safety in dysphagia. As for the latest occurred research trends, fMRI, signals and machine learning emerging into researchers'

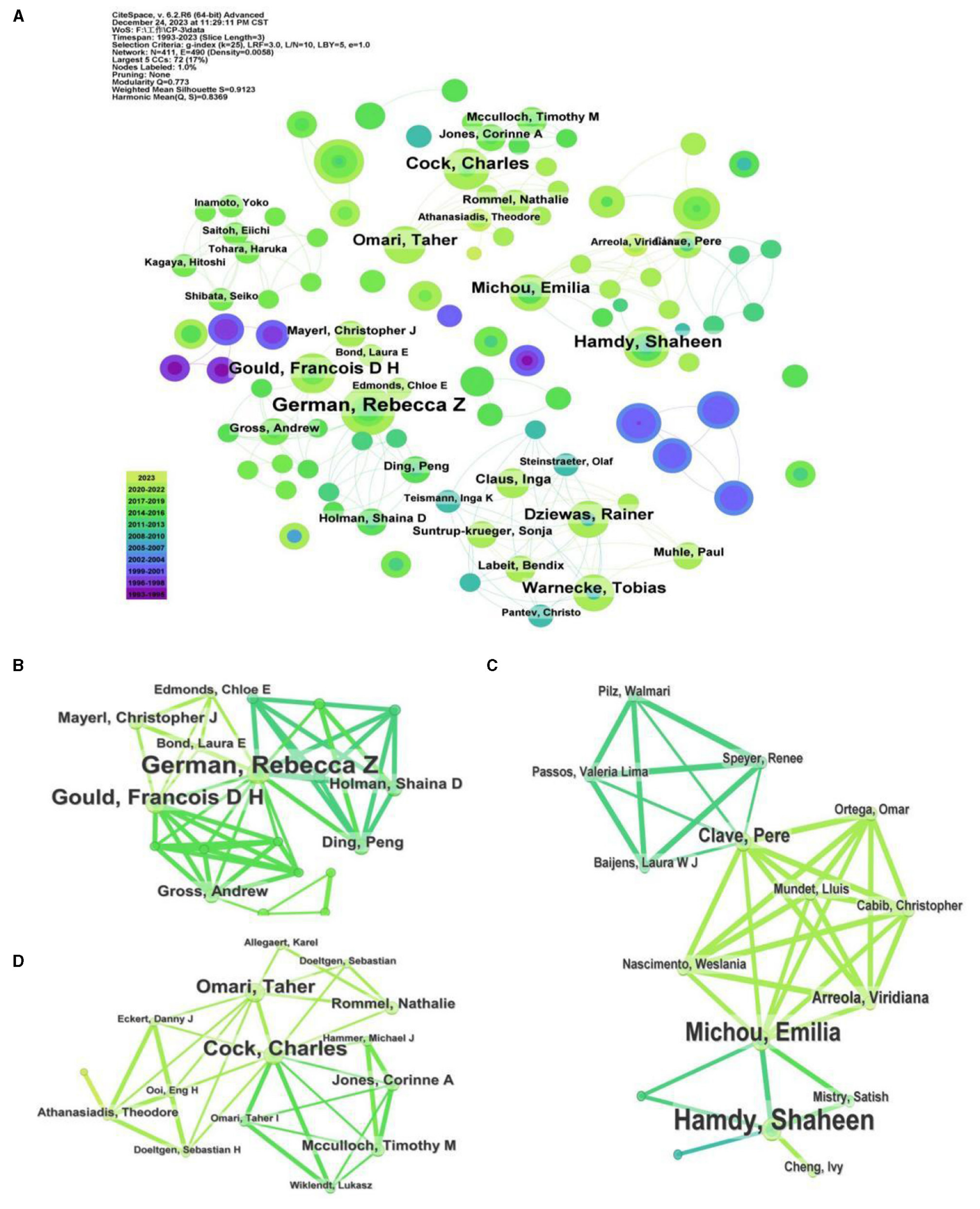

FIGURE 7
(A) Co-occurrence analysis of the authors. **(B–D)** Co-occurrence analysis of the closely work authors.

eyesight. This results showed that collaboration between nations, institutions and regions is uncommon and has to be enhanced going forward. Here, we propose that increased collaboration between specialists from many fields and nations is necessary to implement high-quality multicenter research.

TABLE 5 The top 10 authors in the co-occurrence analysis.

Rank	Author	Documents	Citations	Average citation
1	German, Rebecca Z	14	198	17
2	Jadcherla, Sudarshan R	12	146	13
3	Cock, Charles	10	105	11
4	Steele, Catriona M	10	215	23
5	Hamdy, Shaheen	10	284	36
6	Gould, Francois D H	9	108	15
7	Aydogdu, I	9	625	69
8	Michou, Emilia	8	279	62
9	Warnecke, Tobias	8	159	23
10	Ertekin, C	8	334	42

The study offers insights into the mechanism behind dysphagia from the perspective of visualization approach, which could assist researchers and practitioners in comprehending more comprehensive of dysphagia.

Author contributions

QY: Conceptualization, Funding acquisition, Software, Writing – original draft, Formal analysis. JH: Data curation, Investigation, Methodology, Writing – review & editing. YD: Data curation, Project administration, Validation, Writing – review & editing. HW: Supervision, Visualization, Writing – review & editing. ZD: Resources, Visualization, Writing – review & editing, Supervision.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Cortical compensation mechanism for swallowing recovery in patients with medullary infarction-induced dysphagia

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Introduction: This study aims to examine brain activity during different swallowing actions in patients with dysphagia caused by medullary infarction (MI) before and after treatment using blood oxygen level-dependent (BOLD) functional magnetic resonance imaging.

Methods: Fifteen patients were enrolled in this study. Brain activation during saliva swallowing and effortful saliva swallowing was observed using BOLD imaging in the acute phase of stroke and after 4 weeks of rehabilitation training. Differences in the activation of brain regions during saliva swallowing before and after treatment, during effortful saliva swallowing before and after treatment, and between the two swallowing actions before and after treatment were compared.

Results: In the acute phase of stroke, only the bilateral precentral and left lingual gyrus were partially activated during saliva swallowing, and there was no obvious activation in the insula. Effortful saliva swallowing activated more brain regions than saliva swallowing before treatment, including the bilateral supplementary motor area (SMA), postcentral gyrus, and right insular cortex. The number of brain regions activated during saliva swallowing increased after treatment, including the bilateral precentral gyrus, postcentral gyrus, insula, thalamus, and SMA.

Discussion: Cortical activation increases after recovery from dysphagia, and the increased activation of the postcentral gyrus might play a functional compensatory role. Effortful saliva swallowing is a more effective rehabilitation training method for patients with dysphagia caused by MI.

KEYWORDS

dysphagia, medullary infarction, fMRI, swallowing rehabilitation, FEES, saliva swallowing

1 Introduction

Dysphagia is a common manifestation in stroke patients, occurring in ~25–45% of acute stroke cases, and severe dysphagia is more likely to occur in patients with medullary infarction (MI) (1). Dysphagia can result in malnutrition, increase the risk of aspiration pneumonia, hinder the recovery of patients with stroke, prolong hospitalization, and increase the incidence of adverse outcomes and mortality. Early detection and treatment of dysphagia can reduce subsequent complications, shorten hospital stays, and improve prognosis.

Blood oxygen level-dependent (BOLD) functional magnetic resonance imaging (fMRI) is based on the difference in paramagnetism between oxygenated and deoxygenated hemoglobin and the principle of increased oxygen demand during neuronal activity. This technique reflects the changes in brain activity in various functional brain regions. Owing to the efforts of many experts, BOLD technology has developed rapidly in the past 30 years (2). BOLD-fMRI studies have demonstrated that the cortical regions related to swallowing in healthy individuals include the precentral gyrus, postcentral gyrus, supplementary motor area (SMA), anterior cingulate cortex, insula, frontal gyrus, temporal gyrus, and cuneus, and the activated brain regions differ depending on the specific swallowing actions or types of food (3–6).

In patients with dysphagia after stroke, the brain regions that are activated during swallowing movements change. Because fMRI scanning requires the swallowing action to be performed in the supine position, thereby increasing the risk of choking during swallowing in patients with dysphagia, there are relatively few BOLD imaging studies on patients with dysphagia. Dysphagia can be caused by damage to the brainstem, cortex, and subcortical structures, which result in similar clinical manifestations. At present, current research findings on the activated regions and compensatory mechanisms during swallowing after stroke lack consistency. This may be because the studies failed to differentiate the damaged site or type of swallowing disorder. We aimed to select patients with dorsolateral medullary infarction whose clinical manifestations and Fiberoptic Endoscopic Evaluation of Swallowing (FEES) results were consistent with those of pharyngeal phase dysphagia to minimize variability between participants in the group and obtain more consistent conclusions.

Swallowing rehabilitation is a pivotal intervention for treating dysphagia. However, the cortical compensatory mechanism of rehabilitation training for improving swallowing function and which rehabilitation method is better for specific patients remain uncertain. Therefore, this study aimed to investigate the cortical activation during saliva swallowing and effortful saliva swallowing using BOLD imaging in patients with dysphagia shortly after MI and after dysphagia recovery to explore the cortical compensatory mechanism of dysphagia recovery.

2 Materials and methods

2.1 Participants

We recruited patients with acute stroke of the dorsolateral medulla who met the following criteria and were hospitalized at the Department of Neurology of Tianjin Huanhu Hospital between January 2022 and June 2022. The inclusion criteria were as follows: (i) first-time stroke with confirmed lesions involving the dorsolateral medulla on MRI within 48–72 h of onset; (ii) age between 30 and 75 years; (iii) no history of swallowing disorders before stroke but presenting with dysphagia after stroke, with a score of 3–4 on the Kubota Water Swallowing Test; (iv) clear consciousness, a Mini-Mental State Examination (MMSE) score >24, stable vital signs, and ability to cooperate with the functional examination and swallowing rehabilitation training confirmed by simulated measurements; and (v) voluntarily participated in the study and provided written informed consent.

The exclusion criteria were as follows: (i) dysphagia caused by other neurological diseases or organic lesions; (ii) concomitant cerebral infarction in other brain regions, including the cortex, thalamus, and cerebellum; (iii) patients with metal or other implants who could not undergo 3.0 T MRI; (iv) patients with lesions in the oral cavity, nasal cavity, pharynx, and larynx who could not undergo FEES; and (v) patients with obvious hiccups or severe dysphagia who could not complete the supine swallowing action and those with unstable conditions who were not able to undergo fMRI.

Fifteen patients with an age range of 35–75 years and average age of 54 years were included in the study, and three patients were female. Based on the Edinburgh Handedness Inventory (7), all 15 patients were all right handed before stroke onset (Table 1).

This study was conducted in accordance with the Declaration of Helsinki. The studies involving human participants were reviewed and approved by the Ethics Committee of Tianjin Huanhu Hospital, with approval number (Jinhuan) No. (2020-55). The patients provided their written informed consent to participate in this study.

2.2 Swallowing function evaluation, FEES, and rehabilitation training methods

FEES and BOLD scanning were performed after enrollment, and swallowing rehabilitation training was provided 5 days a week. Swallowing function evaluation, FEES, and BOLD scanning were performed again after 4 weeks of treatment.

2.2.1 Swallowing function evaluation

Swallowing function was assessed using the Kubota Water Swallowing Test (8), wherein the participants drank 30 mL of warm water while seated. The classification criteria were as follows: Grade 1, smooth swallowing of water once; Grade 2, swallowed in two parts without choking; Grade 3, swallowed at once but experienced choking; Grade 4, swallowed in more than two parts with choking; and Grade 5, frequent coughing and inability to swallow.

2.2.2 FEES method

The patients were seated, and topical anesthesia (1% lidocaine + 1/200,000 adrenaline solution) was administered. A fiberoptic endoscope was passed along the floor of the nose through the velopharyngeal port into the pharynx, and the anatomy of the nasopharynx, tongue base, hypopharynx, larynx, and vocal cords, as well as the accumulation of secretions, were observed. We tested the sensation in the throat when the probe touched the arytenoid cartilage. We asked the patient to phonate a high-pitched "eee" to observe the contraction of the pharyngeal muscle and elevation of the larynx. The endoscope was then passed into the hypopharynx, and the patients were asked to swallow fluid (orange juice), semi-solid food (plain yogurt), and solid food (soft bread) in turn to assess leakage, penetration, retention, aspiration, and choking.

TABLE 1 Patient characteristics.

Patient	Age	Gender	Time after (stroke hours)	Lesion side	Score of water swallowing test at enrollment	Score of water swallowing test after treatment
1	75	M	50	L	4	2
2	37	M	52	L	3	1
3	35	M	53	L	3	1
4	65	F	52	R	4	1
5	48	F	53	L	4	2
6	70	M	56	R	3	1
7	48	M	60	R	4	1
8	51	M	57	R	4	2
9	54	M	53	L	3	1
10	55	F	56	R	3	1
11	60	M	60	R	4	2
12	52	M	65	L	3	1
13	59	M	66	R	3	1
14	50	M	60	L	3	1
15	51	M	67	R	3	1

Patient characteristics including age, gender, time after stroke at the first fMRI study (hours), lesion side, and score of Kubota Water Swallowing Test.

2.2.3 Swallowing rehabilitation training

In addition to conventional stroke treatment, various swallowing rehabilitation training methods, including biological low-frequency electrical stimulation of swallowing, cold stimulation training, saliva swallowing training (dry swallow, saliva swallowing action during BOLD scanning) (8), Masako training (effortful saliva swallowing during BOLD scanning, wherein participants squeeze the muscles of the back of their tongue and swallow his saliva as hard as they can) (8), Shaker exercises, and the Mendelsohn maneuver. These methods were utilized to increase sensory stimulation of the pharynx and tongue root strength, improve the contractile force of the pharyngeal muscle, increase the opening time and width of the cricopharyngeal muscle, increase the opening of the upper esophageal sphincter, and improve the sensory and motor function during swallowing (8). Considering the need for a supine examination, two training methods—saliva swallowing and effortful saliva swallowing—were selected for BOLD scanning.

2.3 Magnetic resonance imaging

A 3.0 T MR scanner (Magnetom Skyra, Siemens, Germany) with a 20-channel head coil was used to obtain functional and structural images. Patients were positioned supine on the scanner bed, with their heads fixed in a birdcage-shaped coil, and they viewed the experimental tasks on a screen through a mirror mounted on the head coil. Foam earplugs and pads were used to reduce scanner noise and head motion, respectively. High-resolution T1-weighted structural imaging, functional imaging, and diffusion-weighted imaging (DWI) were performed sequentially, with the scanning plane parallel to the anterior-posterior commissure line. The structural (T1) image parameters were as follows: TR, 2,000 ms; TE, 2.98 ms; matrix, 256 × 256; field of view, 256 × 256 mm; slice thickness, 1 mm; and 192 slices. DWI was performed using the following parameters: TR, 5,200 ms; TE, 80 ms; field of view, 240 mm × 240 mm; 36 slices; and slice thickness, 4 mm. Functional imaging was performed using the magnetic-sensitive GRE-EPI BOLD contrast T2*WI imaging sequence, with the following parameters: TR, 2,000 ms; TE, 30 ms; excitation time, 1; flip angle 90°; field of view, 220 mm × 220 mm; matrix, 64 × 64; 36 slices; slice thickness, 4 mm; no slice gap; and imaging time, 370 s; a total of 90 dynamics were acquired to cover the entire frontal lobe cortex to the level of the medulla oblongata.

2.4 BOLD paradigm

BOLD paradigm is shown in Figure 1. An initial blank resting module of 18 s was set to eliminate the influence of blood flow signal saturation and the magnetic environment. A final blank module of 10 s was set to avoid the final effect of nuclear magnetism. The task employed a BLOCK experimental design, with saliva swallowing and effortful saliva swallowing presented in a pseudorandom manner through visual text prompts. The participants were instructed to perform as many swallowing motions as possible during the swallowing cue period. During the black screen period, the participants rested quietly without any other movements except for breathing. Figure 1 presents the specific task sequence, with each set of swallowing actions lasting for 18 s, followed by an 18 s rest period for a total of 10 sets. The final 10 s blank module was also included, resulting in a total time

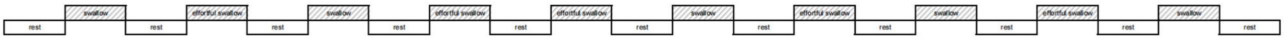

FIGURE 1
Specific task sequence of swallowing during BOLD scanning, each block lasts 18s.

of 370 s. During BOLD scanning, the experimenters accompanied the participants to the MRI room and observed their swallowing actions to ensure that they followed the visual cue requirements for swallowing and resting.

2.5 BOLD image processing

All fMRI data were preprocessed using SPM 12 (Statistical Parametric Mapping, Wellcome Department of Imaging Neuroscience, London, UK) running on MATLAB (MathWorks, Natick, MA). The first 10 time points were removed to eliminate the possibility of unstable AMRI signals. If a patient's motion and rotation parameters exceeded 1.5 mm and 1.5°, respectively, this run of data was excluded from future analysis. The images were subsequently spatially normalized to the Montreal Neurological Institute template brain, resampling voxel size $= 3 \times 3 \times 3$ mm^3. Functional images were spatially smoothed with a three-dimensional Gaussian kernel of 6 mm full width at half maximum to increase the signal-to-noise ratio and reduce inter-subject differences.

2.6 Statistical analysis

The experimental data were organized into a database using Excel, and the SPSS statistical software package version 20.0 (SPSS Inc., Chicago, IL, USA) was used to analyze the activation volume and intensity of the brain functional areas. The General Linear Model (GLM) was used in the first-level analysis.The activated brain regions and their voxels in each participant were tested using one sample *t*-test, To observe the activation in each action, we defined saliva swallowing vs. rest period as con1, effortful saliva swallowing vs. rest period as con2, effortful saliva swallowing minus saliva swallowing as con3. In the second-level analysis, the statistical analysis, the beta weights of two contrasts were statistically compared separately using one sample *t*-test, with the age and sex of participants as covariate. The results were corrected for multiple comparisons using the false discovery rate (FDR) correction at the voxel-level, and the significance threshold was set at FDR-corrected $P < 0.01$ with a minimum cluster size (the number of voxels) of 10 voxels.

3 Results

3.1 General information and FEES results

Table 1 provides an overview of the clinical and demographical characteristics of our patients. All lesions were located on the left or right side of the dorsolateral medulla and were confirmed using MRI (Figure 2). After enrollment, DWI scans were conducted along with two BOLD scans before and after rehabilitation, and no new lesions were observed in comparison to the DWI images prior to inclusion in the study. The score of Kubota Water Swallowing Test after rehabilitation training (1.267 ± 0.4577) was significant lower than that at enrollment (3.400 ± 0.5071; $P < 0.001$, Table 1).

FEES was performed after enrollment (2–3 days after the onset of stroke), revealing impaired pharyngeal swallowing function, indicated by vocal cord paralysis, valleculae and pyriform fossa retention of a saliva-like substance, absence of swallowing reflex upon touching the epiglottis cartilage and posterior pharyngeal wall with a touch stick, a delayed swallowing response, residual vallecular material observed when swallowing viscous food and bread, and leakage and aspiration in some patients. The residual material was removed after repeated swallowing.

After rehabilitation training for 4 weeks, five patients declined follow-up FEES due to their perception of swallowing without any difficulty, and 10 patients underwent a follow-up FEES. Examination of the 10 patients revealed significantly improved pharyngeal swallowing function; there was no vocal cord paralysis and no obvious retention of saliva-like substances in the valleculae or pyriform fossa. In some patients, the swallowing reflex was still delayed upon touching the epiglottis cartilage and posterior pharyngeal wall with a touch stick. Notably, leakage or aspiration was not observed.

3.2 BOLD results for different swallowing actions before and after treatment

3.2.1 Brain regions activated during saliva swallowing in the acute stroke period

After acute stroke, less activation of the brain regions was observed during saliva swallowing, and only the left lingual gyrus and bilateral precentral gyrus were activated. Low-intensity activation was observed in the right inferior occipital lobule, right inferior semilunar lobule, and right posterior lobe of the cerebellum, as shown in Figure 3 and Table 2.

3.2.2 Brain regions activated during effortful saliva swallowing in the acute stroke period

In the acute stroke period, more brain regions were activated during effortful saliva swallowing, including the left lingual gyrus, bilateral precentral gyrus, postcentral gyrus, and SMA, along with low-intensity activation in the right insula, bilateral posterior cerebellum, right inferior temporal gyrus, as shown in Table 3.

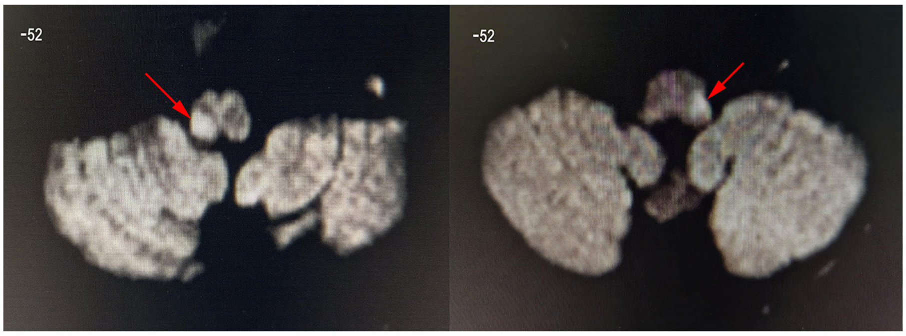

FIGURE 2
Diffusion-weighted images showing dorsolateral medulla infarction (arrow). These are the DWI images of patient 8 (left image, lesion located in the right medulla oblongata) and patient 1 (right image, lesion located in the left medulla oblongata). The z-value of the coordinate for the slice position is −52. All lesions were located in the left or right side of dorsolateral medulla oblongata.

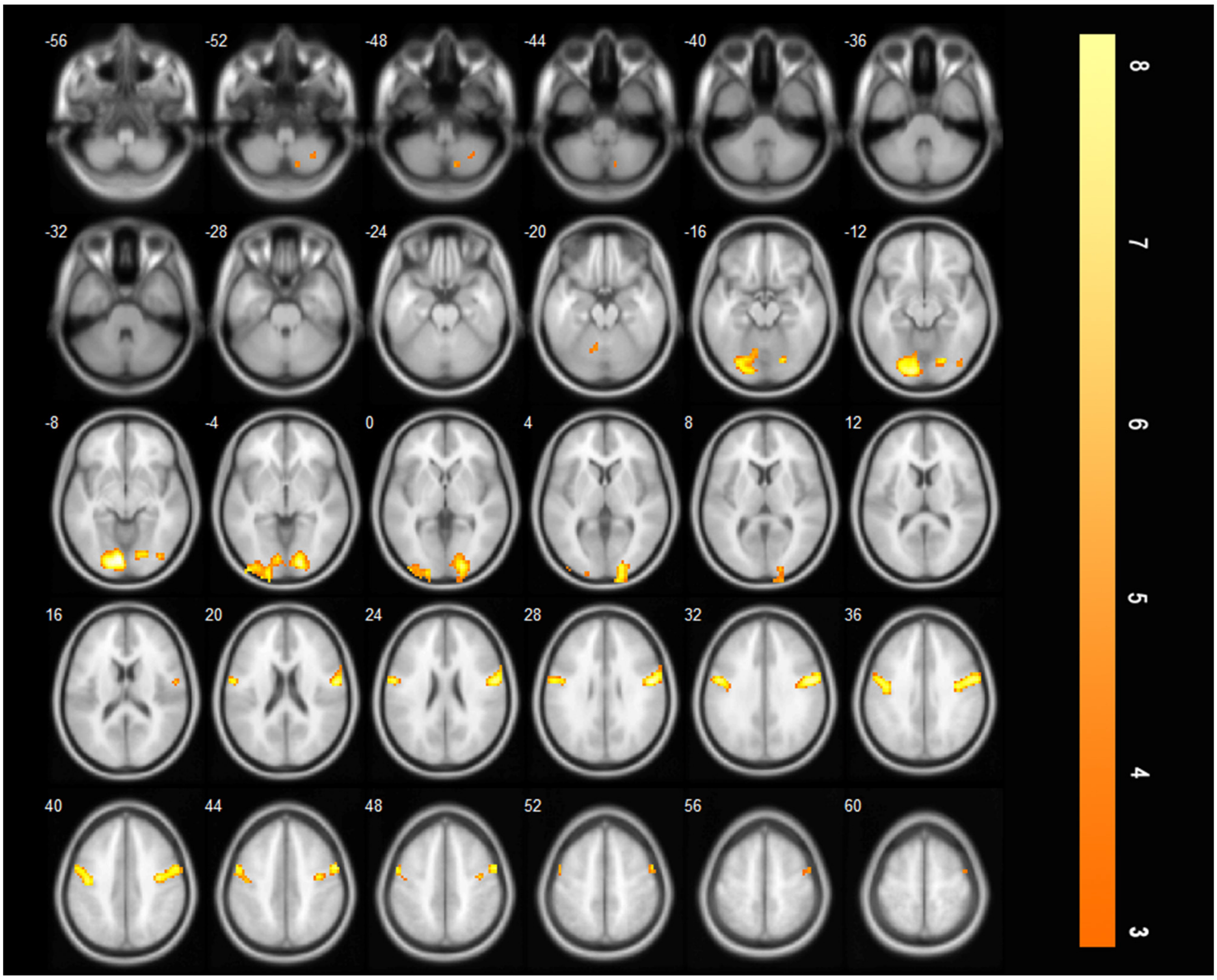

FIGURE 3
Only the left lingual gyrus and bilateral precentral gyrus were activated during saliva swallowing in the acute stroke period ($P < 0.01$, FDR corrected, cluster size > 10 voxels).

TABLE 2 Brain regions activated during saliva swallowing in the acute stroke period.

Regions (AAL)	Cluster size	Peak *T*-value	MNI coordinate		
			X	Y	Z
Lingual _L	321	7.8231	−16	−54	4
Precentral_R	267	7.2283	39	3	45
Precentral_L	208	6.3082	−36	6	45
Occipital Inf_R	19	4.5953	24	−92	−6
Cerebellum 3_R	14	3.5837	8	−42	−18

Cluster size, the number of voxels; MNI, Montreal Neurological Institute; R, right; L, left; $P < 0.01$, FDR corrected, cluster size > 10 voxels.

TABLE 3 Brain regions activated during effortful saliva swallowing in the acute stroke period.

Regions(AAL)	Cluster size	Peak T-value	MNI coordinate		
			X	Y	Z
Cerebellum 3_R	40	5.1808	8	−42	−18
Cerebellum 3_L	39	4.7408	−8	−42	−18
Fusiform_R	56	5.5977	30	−38	−10
Lingual_L	480	7.5498	−16	−54	4
Occipital_Inf_R	16	4.1862	24	−92	−6
Precentral_R	445	9.1773	39	3	45
Postcentral_R	190	4.3643	54	−21	36
Insula_R	12	4.0366	34	16	10
Precentral_L	330	8.4933	−36	6	45
Postcentral_L	219	6.4920	−54	−18	36
Supp_Motor_Area_L	145	6.2439	−6	12	60
Supp_Motor_Area_R	13	4.4967	6	12	60

Cluster size, the number of voxels; MNI, Montreal Neurological Institute; R, right; L, left; $P < 0.01$, FDR corrected, cluster size > 10 voxels.

3.2.3 More brain regions were activated during effortful saliva swallowing than during saliva swallowing in the acute stroke period

The additionally activated regions during effortful saliva swallowing (effortful saliva swallowing minus saliva swallowing) included the bilateral SMA, postcentral gyrus, and right insula, with increased voxel numbers in the bilateral precentral gyrus compared with those regions activated with saliva swallowing in the acute stroke period (Figure 4).

3.2.4 Brain regions activated during saliva swallowing after treatment

After treatment, more brain regions were activated during saliva swallowing, including the left lingual gyrus, bilateral precentral and postcentral gyrus, SMA, insula, thalamus, and low-intensity activation in left inferior frontal gyrus, left calcarine, which are similar to the brain regions activated during saliva swallowing in healthy individuals, as shown in Table 4.

3.2.5 Brain regions activated during effortful saliva swallowing after treatment

After treatment, there are many brain areas activated during effortful saliva swallowing, including the right cerebellum, right superior temporal gyrus, bilateral precentral and postcentral gyri, SMA, insula, thalamus, and bilateral supramarginal gyrus. These regions were similar to those activated in healthy individuals during effortful saliva swallowing (Figure 5).

3.2.6 More brain regions were activated during saliva swallowing after treatment compared with the acute stroke period

Compared with those before treatment, the bilateral precentral and postcentral gyri, bilateral SMA, bilateral insula, and bilateral thalamus were additionally activated during saliva swallowing after rehabilitation training (saliva swallowing after treatment minus that at acute period, Figure 6).

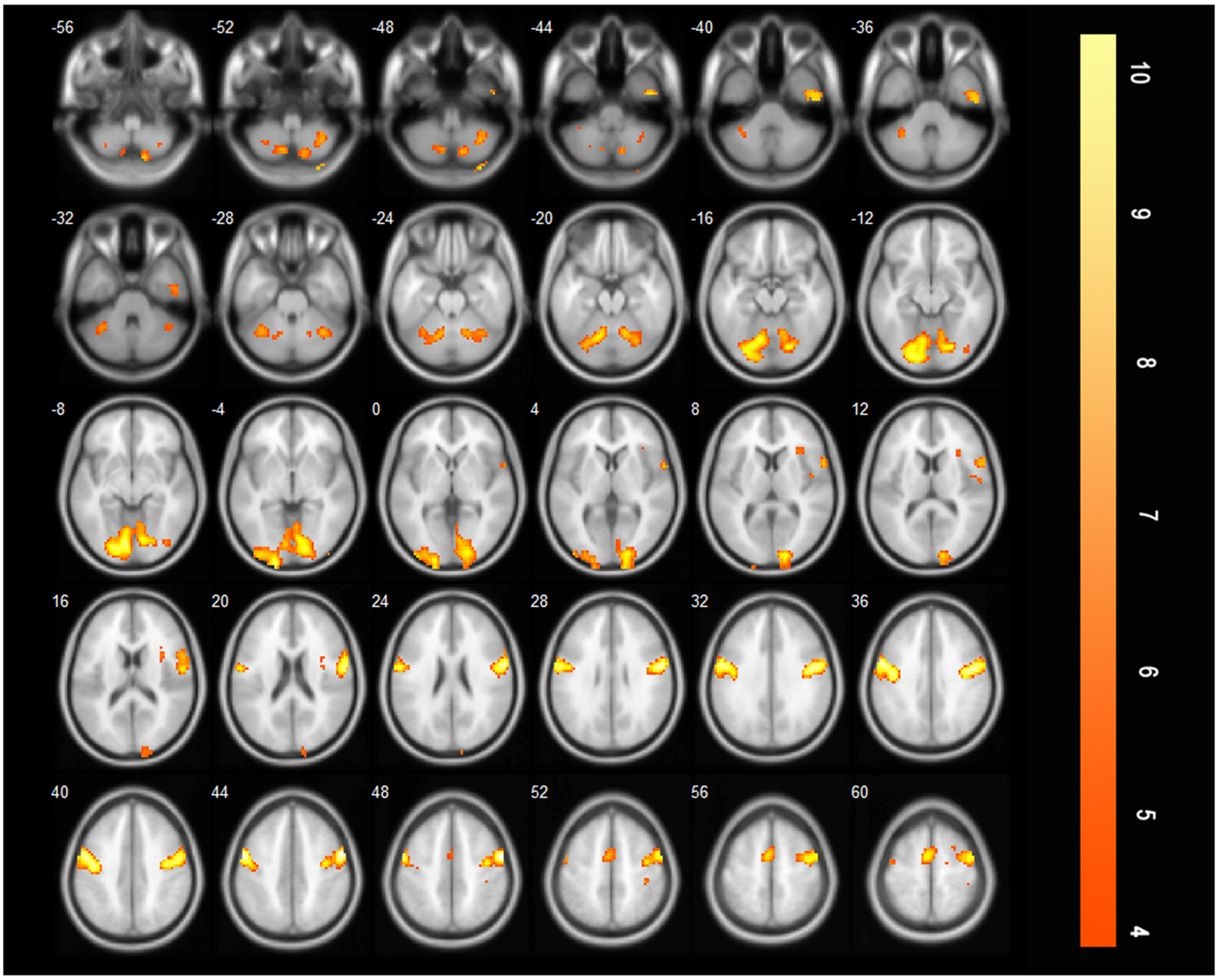

FIGURE 4
T-score map showing difference between two swallowing actions (effortful saliva swallowing minus saliva swallowing) in the acute stage of stroke. Activation of the bilateral SMA, postcentral gyrus, right insula, and the bilateral precentral gyrus can be seen ($P < 0.01$, FDR corrected, cluster size > 10 voxels).

4 Discussion

In this study, we investigated cortical activation during saliva swallowing and effortful saliva swallowing using BOLD imaging shortly after MI and after dysphagia recovery. All patients in our study had dorsolateral medulla damage and relatively severe swallowing difficulties, and some had indwelling gastric tubes. In these patients, swallowing water may increase the risk of choking; therefore, saliva swallowing was chosen as the BOLD paradigm. Some studies have used single-swallow saliva designs for their experiments (4); however, the period was too short for some patients to complete a swallow. Thus, the present study examined continuous saliva swallowing over a longer period. In addition, some experiments evaluated continuous saliva swallowing for 30 s (9). However, in our preliminary tests, we observed that after several consecutive swallows, there was little saliva left to swallow, and additional actions may be required to produce more saliva, which may cause a change in the activated areas. Therefore, an 18 s design was selected, allowing participants to have enough time to rest and produce more saliva.

4.1 Activated regions during saliva swallowing in the acute phase of stroke and insula activation

In this study, very few brain regions were activated during saliva swallowing in the acute phase of stroke. Specifically, only the bilateral precentral gyri and left lingual gyrus were partially activated. This is similar to the findings of previous studies (9), and the right inferior semilunar lobule and right posterior lobe of the cerebellum were slightly activated. The precentral gyrus is the activated cortex for voluntary movement, whereas the lingual and inferior occipital gyri form a visual association cortex that may have been associated with the visual swallowing cues in this study. The inferior semilunar lobule, a new lobe of

TABLE 4 Brain regions activated during saliva swallowing after treatment.

Regions(AAL)	Cluster size	Peak T-value	MNI coordinate		
			X	*Y*	*Z*
Lingual_L	340	7.4724	−16	−54	4
Occipital_Inf_R	19	4.5953	24	−92	−6
Cerebellum 3_R	14	3.5837	8	−42	−18
Frontal_inf_R	35	3.7695	26	36	−16
Calcarine_L	42	5.1877	−10	−92	4
Insula_R	53	5.3279	34	16	10
Thalamus_L	15	4.2452	−14	−18	8
Parahippocampal_L	22	4.0066	−24	−8	−32
Insula_L	27	5.1806	−34	10	10
Precentral_R	579	11.0163	39	3	45
Postcentral_R	182	4.8112	56	−21	36
Precentral_L	479	12.7845	−36	6	45
Postcentral_L	413	6.7225	−63	−9	30
Thalamus_R	21	5.1329	18	−18	8
Supp_Motor_Area_L	72	3.5691	−6	12	72
Supp_Motor_Area_R	76	5.0964	9	12	72

Cluster size, the number of voxels; MNI, Montreal Neurological Institute; R, right; L, left; $P < 0.01$, FDR corrected, cluster size > 10 voxels.

the cerebellum located in the posterior lobe of the cerebellum, receives cortical pontine fibers and exhibits very little activation, which may be related to the coordinated movement of swallowing actions. Although the patients only had a bulbar injury, the small number of activated brain areas indicates that the bulbar region is an important component of the swallowing network (10, 11) and that it influences the related brain regions of the swallowing network. Notably, there was a lack of general cortical activation during swallowing.

Currently, only a few studies have examined brain activation patterns in patients with post-stroke dysphagia. Some studies have shown a significant decrease in the cortical activation areas (9, 12, 13); however, Li et al. reported increased activation in swallowing-related regions, including the left precentral gyrus, left postcentral gyrus, left SMA, and insula (14). Further, Zhou et al. observed a significant reduction in the activation of the anterior cingulate gyrus and excessive activation in other non-swallowing-related regions, including the posterior cingulate gyrus, parahippocampal cortex, visual center, and primary auditory center (15). These inconsistent results may be due to differences between the selected participants. For example, mild-to-moderate dysphagia caused by unilateral hemisphere stroke can produce compensation in the unaffected side or other brain regions (14, 15). However, in the present study, all the patients experienced medullary damage, which is a crucial component of the swallowing network. The medulla participants pharyngeal swallowing and requires bilateral coordination, making functional compensation challenging. Consequently, most cerebral regions cannot be activated, and severe dysphagia is clinically observed. In this study, the patients were in the acute phase of stroke, and cortical activation was scarce, similar to patients with brainstem damage 1–12 months after stroke (9). It is speculated that the activation of the cortex during swallowing in patients with dysphagia is more related to the damaged region and degree than to the post-injury period. If the brainstem damage does not recover, most cortices will exhibit decreased activation regardless of the time after injury.

To date, the conclusions of various studies regarding insular activation in patients with dysphagia have been inconsistent. Patients with unilateral cortical and subcortical infarctions and mild dysphagia exhibited bilateral insular activation (14, 15) with increased activation of the undamaged insula compared with patients in the control group (14), whereas other studies reported no insular activation (9, 12, 13). In this study, all patients had pharyngeal dysphagia, and no insular activation was observed in the acute phase; however, bilateral insular activation was observed after recovery. This finding suggests that the impairment of insular activation may be related to pharyngeal dysphagia. The insula contributes to the initiation of swallowing by processing sensory elements and interacting with widespread brain regions in both hemispheres, including the primary sensorimotor cortex (16). The frontal insula is believed to regulate the temporal organization of mouth movements, such as the timing of the onset of swallowing after a series of chewing or other mouth movements (5). The absence of insular activation may affect the patient's ability to plan swallowing movements, leading to uncoordinated or abnormal swallowing completion, which is manifested as a delay in the start of pharyngeal swallowing. Activation of the insula maybe a compensatory mechanism for the recovery of swallowing function.

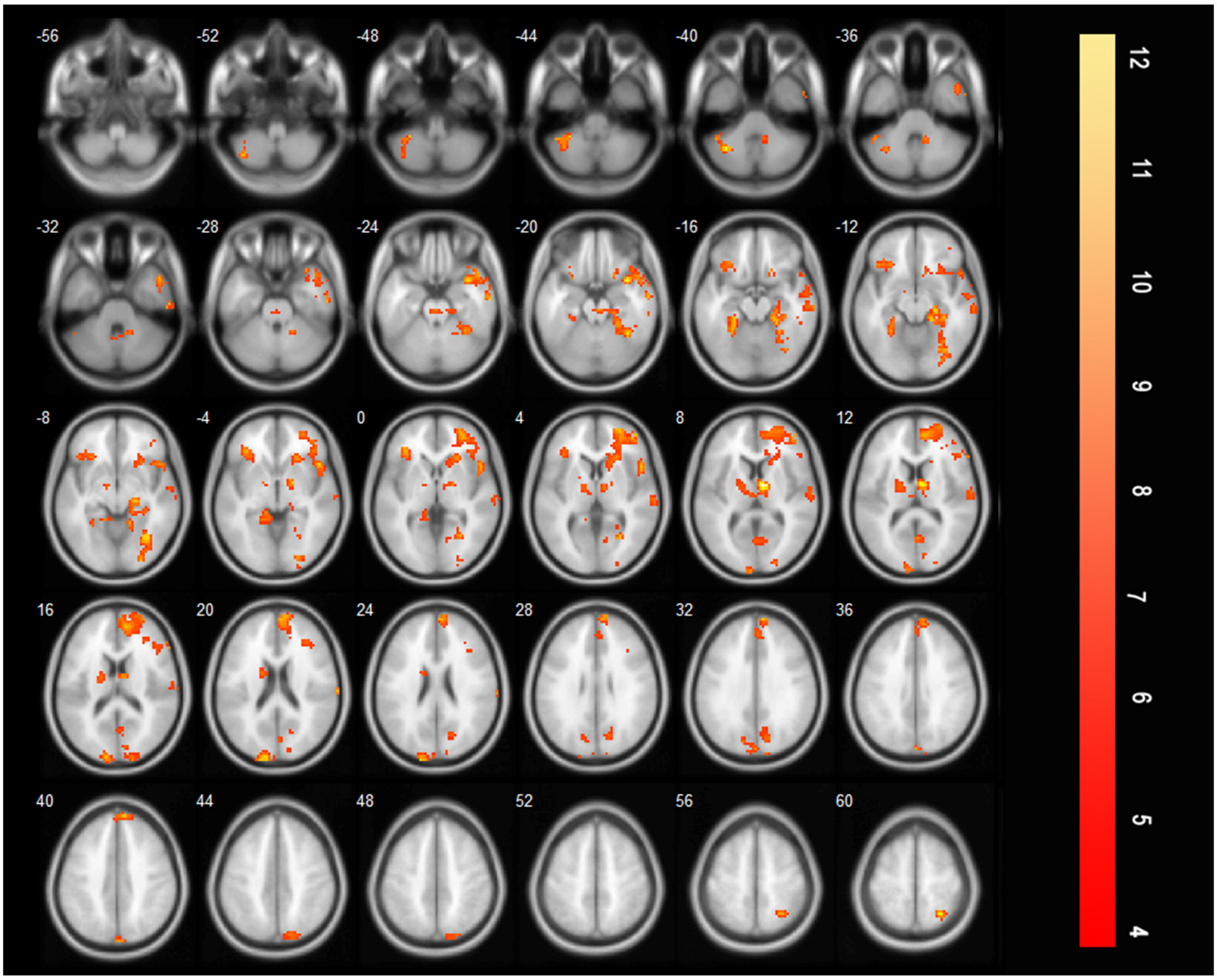

FIGURE 5
T-score map showing many brain areas activated during effortful saliva swallowing after treatment in axial slices, including bilateral precentral and postcentral gyrus, SMA, insula, thalamus, and bilateral supramarginal gyrus ($P < 0.01$, FDR corrected, cluster size > 10 voxels).

4.2 Increased activation of brain regions during swallowing after treatment

Only a few studies have compared the activated brain areas during swallowing before and after rehabilitation. Wei et al. found that the volume of activation in the affected hemisphere increased significantly after rehabilitation, whereas there was no change or only a slight increase in the unaffected hemisphere. Therefore, the recovery of cerebral hemisphere function on the affected side is important for the rehabilitation of patients with swallowing disorders (13). In contrast, Li et al. observed an increase in activation in the contralateral hemisphere during the acute phase of stroke and suggested that compensation from the intact side promoted functional recovery (14). These inconsistent results may be attributed to the heterogeneity of the enrolled patients. To increase accuracy, it is advisable to select relatively consistent patients, such as those with unilateral involvement of a certain cortical or subcortical structure.

After language training, patients with dysphagia caused by unilateral hemisphere stroke exhibited significant activation of the bilateral primary motor areas, sensory areas, premotor areas, and the anterior insula during swallowing (6). In patients with brainstem damage, increased activation of brain regions, including the cingulate gyrus, insula, precuneus, frontal lobe, orbitofrontal cortex, and SMAs, was observed after treatment. These findings suggest that other brain regions have some compensatory ability when a swallowing center injury occurs (9). Our study demonstrated the bilateral precentral gyrus, postcentral gyrus, insula, thalamus, and SMA showed increased activation after rehabilitation training. These regions are not completely consistent with the above results (9), which may be attributed to different post-injury periods and different compensatory mechanisms between the early and chronic periods.

Mihai et al. found that even after swallowing function recovery, the overall activation level of swallowing-related brain regions in patients was still lower than that of the control group (17).

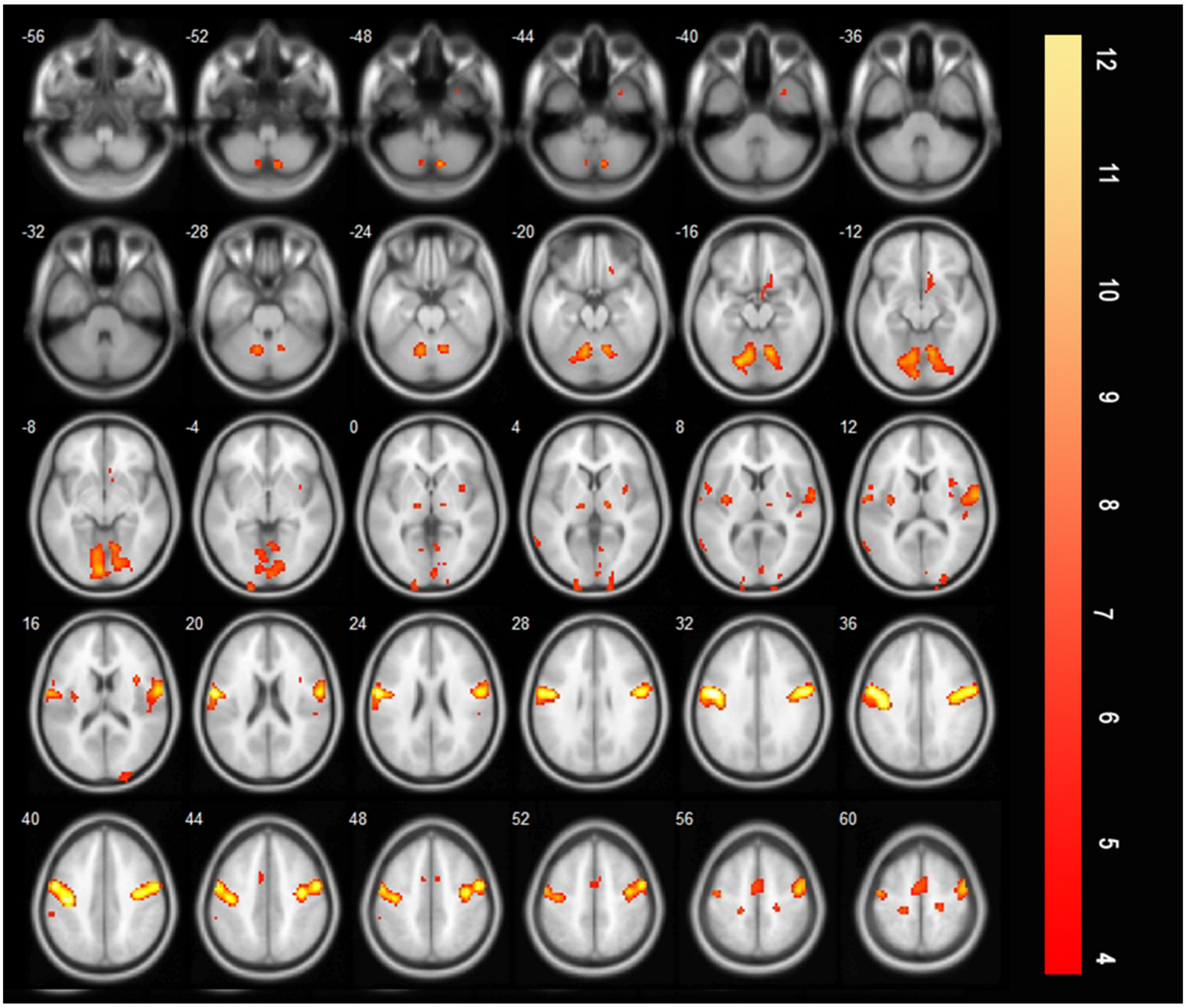

FIGURE 6
T-score map showing the difference during saliva swallowing between two timepoints (saliva swallowing after treatment minus saliva swallowing in the acute stroke period), the bilateral precentral and postcentral gyrus, bilateral SMA, bilateral insula, and bilateral thalamus were additionally activated ($P < 0.01$, FDR corrected, cluster size > 10 voxels).

Meanwhile, there was increased activation in the contralateral S1 area; the more obvious the initial damage, the more obvious the activation in the contralateral S1 area after functional recovery. This indicates that activation of the contralateral S1 area is the reason for the recovery of swallowing function. Domin et al. revealed that the integrity of callosal fibers interconnecting the S1 swallowing representation were significantly associated with swallowing compliance (18). An increase in activation was observed in the postcentral gyrus in this study, which is similar to previous results (17, 18). This suggests that after the motor function is impaired, increased somatosensory input is required to complete swallowing tasks; that is, sensory reorganization contributes to compensation.

Oral sensory stimulation can activate the sensory and motor areas associated with swallowing, which can enhance sensory input and participate in the control center of the brainstem and cortex, potentially affecting the autonomous components of swallowing (19). Studies have revealed that pharyngeal stimulation first activates the sensory-motor cortex, then excites the swallowing motor cortex and changes the recruitment pattern of cortical activation related to swallowing (20, 21). In this study, all patients had pharyngeal dysphagia, and FEES revealed decreased sensation in the pharynx. When the probe touched the epiglottis and posterior pharyngeal wall, it did not elicit a swallowing reflex, causing a delay in the swallowing response; this is an important cause of choking. The patients in this study underwent ice-cotton swab stimulation training, which improved pharyngeal sensation and increased sensory input to the postcentral gyrus, possibly by activating the thalamus. Sensory compensation contributed to clinical recovery, as confirmed using FEES, which showed improved pharyngeal sensation after recovery. This finding suggests that the tactile input during swallowing may be a crucial factor in recovery. In the treatment of dysphagia, especially pharyngeal dysphagia, it is necessary to focus not only on motor training but also on sensory stimulation training to better promote swallowing function.

4.3 Comparison of brain activation between effortful saliva swallowing and saliva swallowing before and after treatment

After treatment, there was only a slight increase in activation in the left SMA, right supramarginal gyrus, and left postcentral gyrus when effortful saliva swallowing was compared with saliva swallowing. Peck et al. found that healthy individuals exhibited increased activation in the angular gyrus, cingulate gyrus, inferior parietal lobule, middle frontal gyrus, superior frontal gyrus, and supramarginal gyrus when comparing effortful saliva swallowing to saliva swallowing (22). The results of the present study were similar, although the range of differences in activation was smaller. However, before treatment, a significant increase was observed in brain activation between the two swallowing actions, including the bilateral SMA, bilateral postcentral gyri, and right insula. This finding suggests that patients with acute medullary stroke need to exert more effort to complete effortful saliva swallowing and activate more brain regions, which are the compensatory areas activated when normal saliva swallowing is performed after recovery.

Gandhi et al. (23) found that effortful swallowing can improve swallowing function in patients with PD more effectively than normal swallowing, whereas Kim et al. (24) found that tongue-to-palate training can increase the oral-pharyngeal swallowing function of subacute stroke patients. The results of these two studies are consistent with our findings. Additionally, Bahia et al. reviewed 23 studies and found that the biomechanical effects of effortful swallowing include increased pressure in the oral, pharyngeal, and esophageal regions (25). The BOLD imaging results confirmed that the recovery of swallowing function after stroke can be attributed to the compensation of more brain regions; however, the results cannot confirm that rehabilitation training itself stimulates the compensatory brain regions. Here, we observed that effortful swallowing after MI can activate these compensatory brain regions and promote faster recovery of swallowing function. This indicates that in pharyngeal dysphagia caused by MI, effortful saliva swallowing can activate more brain regions and better promote the recovery of swallowing function, making it a more suitable rehabilitation training method than saliva swallowing.

5 Study limitations and future directions

This study has some limitations. First, to ensure the comfort of patients during BOLD study, saliva swallowing and effortful saliva swallowing actions were observed subjectively by the experimenters without objective quality control. Nevertheless, documenting the quality control of the performance of participants during fMRI is very important (2). In future studies, electromyography or pads placed on the neck to record the thyroid cartilage motion could be used to mark swallowing. Second, FEES was only used to confirm that the enrolled patients had pharyngeal dysphagia at the initial design of the experiment, so no numerical scoring was performed. However, if FEES scoring was available for the improvement of pharyngeal sensation, it would be more accurate. Third, only 15 patients were enrolled, and the MI was not differentiated between the left and right sides, making it impossible to distinguish whether the contralesional or ipsilesional hemispheres were more compensatory. In the future, increasing the number of trial participants and grouping them according to the injured side can ensure more accurate observation of compensatory mechanisms.

Although the Mendelson maneuver can also be used to increase cortical activation in healthy individuals (22), it requires a 3 s breath hold after lifting the larynx; this could not be accurately controlled in this study. Moreover, breath-holding during MRI may pose a risk for patients with MI. In the future, such training could be examined under the premise of ensuring patient safety. Jing et al. found that observing swallowing movements could activate mirror neurons and swallowing networks in healthy individuals, indicating the potential value of action observation in the treatment of swallowing disorders (26). We can observe whether this safe training technique is useful for patients with dysphagia in the future.

6 Conclusion

This study demonstrated that patients with dysphagia after MI did not exhibit insular activation at the acute stroke period. After recovery, activation of the posterior central gyrus increased, which is consistent with the improved swallowing function and pharyngeal sensation in FEES. The BOLD results confirmed that the sensory cortex might play a compensatory role in the recovery of swallowing function. Simultaneously, effortful saliva swallowing after MI significantly increased activation in swallowing-related regions, and these brain regions promoted the recovery of swallowing function, suggesting that effortful saliva swallowing is a more effective rehabilitation training method for patients with dysphagia after MI.

Data availability statement

The data analyzed in this study is subject to the following licenses/restrictions: The raw data is the result of fMRI, which is saved in the form of a CD and requires software analysis. The figures and tables attached in the article can be used publicly, but only members of the research team can view the original MRI images. Requests to access these datasets should be directed to FG, googledragon@163.com.

Ethics statement

The studies involving humans were approved by Ethics Committee of Tianjin Huanhu Hospital. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

FG: Data curation, Investigation, Methodology, Writing – original draft, Conceptualization, Writing – review & editing. JH: Data curation, Investigation, Writing – original draft. QZ: Data curation, Investigation, Writing – original draft. XL: Methodology, Writing – original draft. YW: Data curation, Investigation, Writing – original draft. JW: Project administration, Supervision, Writing – review & editing, Methodology.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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