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# ORIGINAL ARTICLE

# Efficacy of a Compound Swallowing Training Program Incorporating Neuromuscular Electrical Stimulation on Postoperative Chronic Dysphagia in Patients with Tongue Cancer

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# Abstract

*Objective*: To investigate therapeutic effects of a compound swallowing training program(CSTP) that includes neuromuscular electrical stimulation, swallowing muscle strengthening, posture training, food texture modification, and the use of adaptive feeding devices on chronic dysphagia after surgical treatment of tongue cancer(TC), and review the long-term swallowing outcomes and possible adverse effects.

*Materials and methods*: In this retrospective longitudinal study, we included patients with chronic dysphagia after surgical treatment of TC, who received a total of 10 CSTPs three times weekly, for 1 h each time. Penetration-Aspiration Scale (PAS), Functional Dysphagia Scale (FDS), and Swallowing Performance Rating Scale (SPS) scores were compared before and after curative treatment and at the 6-month follow-up.

*Results*: Twelve patients were included in this study (11 men and one woman; mean age: 56 years; range: 42–74 years). The initial assessment was performed an average of 11.9 months after surgery (range: 6–34 months). There was a significant improvement in the SPS scores one month and six months after completing CSTP (p = 0.015, p = 0.024). Six months after completing CSTP, the total FDS scores of 5 mL of thin liquids and 5 mL of thick liquids significantly improved compared with those before training (p = 0.007, p = 0.011). Three of seven patients requiring chronic tube feeding had the nasogastric tube successfully removed after training.

*Conclusion*: This study suggested that CSTP has positive effect on improving the swallowing function, based on the SPS and FDS score on the liquid swallowing test in TC patients with chronic dysphagia. CSPT may increase the likelihood of feeding tube removal in TC patient with chronic dysphagia.

Keywords: Tongue cancer, Dysphagia, Neuromuscular electrical stimulation, Swallowing training

# 1. Introduction

O ral cancers are a form of malignant head and neck tumors (HNC) that are associated with significant morbidity and mortality, and account for a 4.98–7.25% of all malignant tumors.<sup>1</sup> Tongue cancer (TC) is the one of the most common oral cancers. The proportional rate of TC to all oropharyngeal cancer accounted for 26.4–26.67% in

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men and 28.77-34.85% in women in Taiwan.<sup>2</sup>

More than 50% of patients with isolated tongue resection or tongue resection mixed with anterior floor resection (including partial mandibular, anterior floor, sublingual gland, and neck dissection) have significant swallowing impairments.<sup>3</sup> Removal of the floor of the mouth and the base of the tongue may affect the oral and pharyngeal stages of swallowing.<sup>3,4</sup> Patients with advanced cancer usually require radiation and/or chemotherapy, which often results in reduced salivary secretion, further damage to tiny blood vessels, fibrosis of adjacent muscles and soft tissues, long-term xerostomia, and prolonged food transit time during the oral and pharyngeal phases of swallowing.<sup>4</sup> Dysphagia owing to adverse effects caused by cancer treatments may further lead to malnutrition, dietary restrictions, choking, pneumonia and even life-threatening complications.<sup>4,5</sup>

Traditional swallowing therapy, including the Shaker exercise, Mendelsohn maneuver, supraglottic swallow maneuver, tongueholding maneuver, repeated swallow, effortful swallow, and food texture modification, are promising to improve swallowing function in patients with HNC.4,6 Prophylactic swallowing exercises have been shown to be effective in maintaining pharyngeal muscle tone and improving swallowing function at 3 and 6-month post treatment.<sup>7</sup> Zhang et al.<sup>8</sup> used direct and indirect swallowing therapy in patients with dysphagia after TC surgery and found that it effectively improved postoperative dysphagia and quality of life. Recent studies suggest that motor exercise before and during radiation may reduce chronic dysphagia in patients with advanced HNC.<sup>6–9</sup> Research reported that in 12–69% of patients with HNC, dysphagia did not improve 3-6 months after treatment, and it even got worse in 8-20% of patients.<sup>6,10</sup> Therefore, an effective swallowing treatment protocol should be developed in order to improve the swallowing function of patients with HNC suffering from chronic dysphagia.

Decrease hyolaryngeal excursion caused by muscle weakness can affect swallowing performance and increase the risk of aspiration.<sup>4</sup> Head lifting exercise, focused on strengthening the submental muscles, were suggested effective for pharyngeal dysphagia in HNC.<sup>10</sup> Post exercise surface electromyography demonstrated signs of fatigue by reducing muscle activity. This evidence provides support of swallowingrelated fatigue from exercise.<sup>10</sup> For physically debilitated patients, they may not be able to fully cooperate with these exercise training and the treatment may fail. Therefore, the efficacy for exercise depends on the compliance of the patients.<sup>11</sup> Continual eating over the course of a meal requires endurance. For patients with dysphagia, impaired endurance or fatigue may affect swallowing performance and increases the risk of aspiration.<sup>12</sup> Clinically treatment recommendations include shortening meal time, taking more frequent meals, adjusting food texture, and shortening treatment session duration to minimize fatigue effects.<sup>12</sup> As there is still a lack of clinical guidelines, further study is required to establish a common framework for clinician.

Neuromuscular electrical stimulation (NMES) has been suggested as a potential treatment strategy for oropharyngeal dysphagia.<sup>13–15</sup> When applied to the neck muscles, NMES may lead to more powerful contractions of the swallowing muscles, thus enhancing the hyoid excursion and strength laryngeal elevation, which leads to better swallowing effort.<sup>16</sup> Most early studies have focused on the treatment of neurological dysphagia and how to increase the removal rate of nasogastric and gastrostomy tubes.<sup>13,15</sup> Research has demonstrated that using NMES as a conjunction therapy can improve oropharyngeal function and enable greater oral intake of normal food consistencies, but no relevant effect was found when used alone.<sup>15</sup> Lin et al.<sup>14</sup> studied the use of functional electrical stimulation for the treatment of dysphagia after radiotherapy for nasopharyngeal carcinoma. Compared with home exercises, functional electrical stimulation is superior in terms of the improvement of various indicators of swallowing and quality of daily life. Ryu et al.<sup>15</sup> studied the use of NMES combined with traditional swallowing exercise therapy to treat dysphagia in the acute treatment phase of HNC, and found that its effect was better than that of traditional therapy alone. However, a large, randomized, controlled clinical trial determined that NMES did not add any benefit to traditional dysphagia therapy for post-radiation dysphagia in HNC after 12 weeks of training, and the active NMES group had significantly worse penetration aspiration scale scores than did the sham group.<sup>17</sup> Although NMES has been used as a supplement for swallowing training, its efficacy is still inconsistent. Reference evidence and standard protocols are lacking for the treatment of chronic dysphagia in patients with TC.

A compound swallowing training program (CSTP) that combines NMES, swallowing muscle strengthening exercise, posture adjustment, stepwise food texture modification and instructions on the use of adaptive feeding devices was used to treat dysphagia in patients with HNC. Premising that NMES can be considered as an adjunctive treatment modality for dysphagia, and considering the patients' ability to eat each meal and possible swallowing-related fatigue from exercise therapy in patients with HNC, in this 1- hour training protocol, combined therapy was only performed in second 30 min of the hour, while NMES is performed in the first 30 min. We hypothesized that strengthening the atrophy or fibrotic muscles by using non-active locomotor tools (NMES), followed by active exercise and various swallowing techniques, can also effectively improve swallowing function for chronic severe dysphagia in TC. Therefore, the present study aimed to investigate the efficacy of CSTP for postoperative chronic dysphagia in patients with TC and evaluate its long-term benefits to serve as a reference for clinical treatment and rehabilitation.

# 2. Materials and methods

# 2.1. Participants sampling

This was a retrospective longitudinal study that included patients with chronic dysphagia, who were referred by the HNC team to the rehabilitation department after TC surgery, radiation and/or chemotherapy, from 2013 to 2016. The TC was diagnosed, localized, and staged based on the medical history, physical examination, computed tomography or magnetic resonance imaging, and postoperative pathology reports of the patients. The inclusion criteria were as follows: (1) Patients with single-site TC, had undergone surgery more than 6 months earlier and had received traditional swallowing training, and had a persistent swallowing disorder with a functional oral feeding scale (FOIS) score  $\leq$ 4. Their skin condition at the treatment site was stable, and had no signs of wound, inflammation, or cancer recurrence. (2) Those had finished 10 sessions of CSTP, and had completed three videofluoroscopic swallowing studies (VFSS). We excluded (1) patients with dysphagia caused by other neuromuscular and skeletal lesions that existed before the TC treatment, (2) those aged under 18 years, and (3) those underwent laryngectomy. The participants were selected from the database of our "Head and Neck Cancer Research Proposal" (CMRP101-2397A3). This study was approved by the Institutional Review Broad (IRB) of the Chang Gung Medical Foundation (IRB number: 202201217BO). The IRB approved the waiver of the participants' consent.

# 2.2. Research design

# 2.2.1. Collection of basic participants' data

The following information was recoded for analysis and comparison: age, sex, cancer site, classification, stage, time of occurrence, and date of referral.

#### 2.2.2. Treatment protocol

A CSTP that combines neuromuscular electrical stimulation, swallowing muscle strengthening exercises, posture adjustment (chin-up, chin-down, head rotation, head tilting),<sup>4</sup> stepwise food texture modification, and instructions on the use of adaptive feeding devices (compressible long-mouth bottle, long-arm spoon, syringe, and chopsticks)<sup>4</sup> was performed in this study.

A dual-channel electrical stimulation therapy device (VitalStim TM, Chattanooga Group, Hixon, TN) and a compatible specific electrode patch were used.

This is a 1-h training protocol, involving 30 min of NMES for the swallowing muscles, followed by 30 min of combined therapy of NMES and compound swallowing training. During the first half hour of the process, only NMES was performed, followed by synchronous therapy with oral exercise, compensatory postural training, Mendelsohn maneuver, and stepwise food preparation guidance combined with NMES in the following half hour. The need for adaptive feeding devices was also suggested individualized according to the first swallowing evaluation and the patients were educated to use the device during the process. Treatments treatment were administered three times per week, each session lasted 1 h, and a total of 10 sessions were performed.

NMES adopts a dual-channel synchronous electrical stimulation: the first pair of electrodes was placed horizontally above the hyoid bone, and the second pair of electrodes was placed longitudinally under the hyoid bone and in the recess of the thyroid cartilage. If there was a flap or hypertrophic scar in the area above, the electrodes were applied laterally on the unaffected side (Fig. 1). Placing the second pair of electrodes longitudinally under the hyoid bone may keep away from carotid sinuses on both sides, reducing the risk of carotid sinus massage during electrical stimulation. The electrodes were also placed as close as possible to the thyrohyoid muscle belly to enhance the upward movement of thyroid cartilage.<sup>11,14,18</sup> While monitoring the patient's feelings, the current intensity was gradually increased until the throat became tightened. This was regarded as the current intensity of muscle contraction, and this intensity was maintained for a period of 1 h.<sup>18</sup>

# 2.2.3. Swallowing assessment and scoring method

a. Videofluoroscopic Swallowing Studies (VFSS)

VFSS was performed at three different time points to evaluate various data; namely, before treatment (T0), one week after completing the treatment course (T1), and six months after completing the course of treatment (T2). A retrospective chart review was made five years after the treatment (T3) to investigate the patient's swallowing performance and health status in the chronic phase.

This examination, performed in collaboration with a radiologist and speech therapist, used the lateral and anterior-posterior views to assess swallowing. Each patient used 3 and 5 mL of thin liquid, equivalent to the International Dysphagia Diet Standardisation Initiative (IDDSI) level 1, respectively. If there was no severe difficulty with oral delivery (oral transit time [OTT] greater

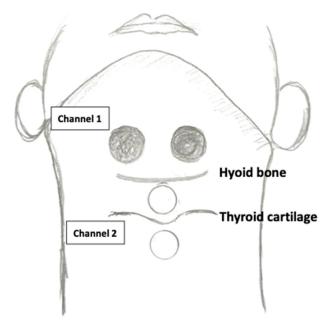


Fig. 1. Electrodes placement.

than 5 s) and no significant aspiration, we proceeded to the next 3 and 5 mL thick liquid (IDDSI level 2) tests and puree (IDDSI level 4) test. The test was terminated once overly prolonged OTT or significant aspiration was observed.<sup>19</sup> In this study, we defined "significant aspiration" as the aspirated bolus dropped into the lower 1/3 of the trachea and/or coated on the trachea wall.

The results were graded by a senior speech therapist experienced in performing VFSS. The presence or absence of penetration and aspiration was defined according to the Penetration-Aspiration Scale (PAS), which is an eight-points scale used to define the severity of choking and aspiration. A score of 2–5 was defined as penetration and a score of 6–8 was defined as aspiration.<sup>20</sup>

# b. Functional Dysphagia Scale (FDS)

Based on the indicators of the video swallowing examination, FDS was used to evaluate the severity of the dysphagia, bolus advancement, oral residues, and transit time. A total of 10 items were used, with the total score ranging from 0 to 100, the higher the score, the more serious the dysphagia. The items included in the scale were lip closure, bolus formation, oral residues (%), oral transit time (OTT), triggering of the pharyngeal swallowing reflex, laryngeal elevation and epiglottis closure, nasal penetration (%), residues in the vallecula and pyriform sinus (%), pharyngeal wall coating after swallowing, and pharyngeal transit time (PTT). Oral residue was defined as any residue in the mouth after swallowing. The OTT was defined as the time interval from the beginning of the movement of the bolus in the mouth until the first (front) part of the bolus enters the pharynx through the intersection of the mandible and the base of the tongue. A delay was defined when the OTT exceeded 1.5 s. PTT was defined as the time interval from the passage of the front of the bolus (through the intersection) to the passage of the tail of the bolus through the upper esophageal sphincter. A delay was defined as a PTT >1 s. Higher scores on this scale indicate worse swallowing performance in the VFSS.<sup>21</sup>

c. Clinical swallowing assessment using the swallowing performance scale (SPS)

This is an assessment tool of swallowing disorders and feeding plan recommendation that combines clinical and VFSS findings. The scale grades swallowing disorders with scores ranging from 0 to 7, with higher scores representing more severe swallowing disorders; a score of 1 corresponds to normal oral intake, meanwhile a score of 7 corresponds to severe impairment with significant aspiration, or insufficient transit from the oropharynx to the esophagus, requiring enteral nutritional support.<sup>22</sup>

# 2.2.4. Statistical methods

The collected data were encoded and input for statistical analysis. Wilcoxon's Signed Rank test was used for the analysis of median changes over time in SPS, PAS and FDS for comparison of the values between different time points during the treatment course.

The results based on continuous variables are presented as median and IQR due to the skewed distribution of most variables. IBM SPSS Statistics version 25 (IBM Corp.) was used for all tests and the level of statistical significance was set at P < 0.05. For multiple comparisons, the Bonferroni correction was applied, and the level of statistical significance was corrected to P < 0.017.

# 3. Results

Twelve patients (11 males and 1 female) with chronic dysphagia secondary to TC surgery were included in this study. The average age of the patients was 56 (range, 42-74) years. Four (33.3%) patients had cancer of the base of the tongue. At the time of participation in the treatment plan, the clinical stage of the patients was ranged from stage II to IV, with 58% among them having stage IV cancer. The histopathological diagnosis was squamous cell carcinoma. All patients underwent surgery and radiation therapy. Among them, six (50%) underwent total tongue resection and eight received mandibulectomy. The initial assessment was performed at 11.9 (range, 6-34) months postoperatively. The basic patient information is listed in Table 1.

Table 1. Basic characteristics of patients.

	Number of people	%
	(n = 12)	
Age(y/o)	$56 \pm 8.13$	
Gender		
Male	11	91.7
Female	1	8.3
BMI	$21.79 \pm 3.14$	
Site		
Oral tongue	9	75
Basal tongue	3	25
Stage		
Т		
1/2/3/4	0/4/1/7	0/33.3/8.3/58.3
Ν		
0/1/2	10/1/1	83.3/8.3/8.3
М		
0/1	12/0	
Resection	12	100/0
Total tongue resection	6	50
2/3 tongue resection	3	25
Wide excision	3	25
Radiation therapy	12	100
Chemotherapy	4	33.3
Time to start treatment (months)	11.8 ± 8.59	

Table 2 shows the results of the VFSS assessment (using liquid barium) before the treatment. All patients had limited tongue movement, more than half had a delayed oral transit time, 83.7% had oral residue, and 75% had varying degrees of difficulties in mouth opening. The main problems in the pharyngeal phase were the presence of residues in the vallecular and pyriform sinus, and 10 patients (83.3%) had penetration or aspiration.

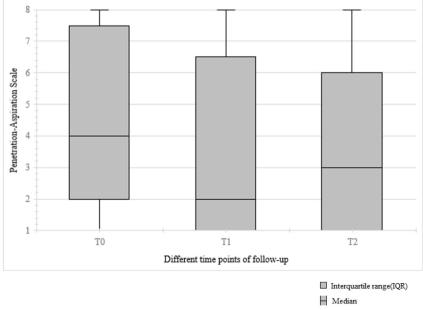
After the intervention of CSTP, swallowing function improved generally, and five patients (41.3%) had improved PAS scores. Five patients had improved PAS scores at T2 compared to the scores at T0. Changes in PAS scores at different time points are shown in Fig. 2. Seven patients (58%) had improved SPS grades after treatment, and four patients (33%) showed further improvements at T2. The changes in the SPS scores at different time points are shown in Fig. 3. There was a significant difference in the SPS grades at T1 and T2 (p = 0.015, p = 0.01). Regarding the VFSS, the FDS scores of different barium bolus textures improved after treatment but not significantly. In the follow up study, the FDS scores of 5 mL thin liquid and 5 mL thick liquid bolus at T2 were significant improved when comparing to those at T0 (p = 0.007, p = 0.011). In the comparison between T2 and T1, we also found a significant improvement in 5 mL thin liquid and 3 mL thick liquid bolus (p = 0.007, p = 0.017). These finding showed that the swallowing function in our patients continued to improve after completing treatment (Table 3).

In 5 years review of the 12 patients after CSTP, three among the seven patients who required long-term tube feeding before treatment could successfully get nourished without the nasogastric tube after the treatment. Two patients developed facial cellulitis during the follow-up period; however, they recovered after antibiotic treatment. After

Patient	Difficulty in mouth opening	Limited tongue movement	Oral residue	Delayed OTT	Nasal penetration	VR	PR	Penetration	Aspiration
1	+	+	+	+	+	+	+	_	_
2	+	+	+	+	+	+	+	+	_
3	+	+	-	_	-	_	_	+	_
4	+	+	+	+	_	+	_	+	—
5	_	+	+	+	_	+	+	+	+
6	+	+	+	+	-	+	_	+	—
7	_	+	+	+	_	+	+	+	+
8	_	+	-	_	-	_	_	+	+
9	+	+	+	_	_	_	_	+	—
10	+	+	+	+	-	+	+	+	+
11	+	+	+	+	_	+	+	+	_
12	+	+	+	+	_	+	+	_	_
N (%)	9(75)	12(100)	10(83.7)	9(75)	2(16.6)	9(75)	7(58.3)	10(83.3)	4(33.3)

Table 2. Results of VFSS assessment of liquid barium before treatment.

OTT: oral transit time, VR: vallecular residue, PR: Pyriform sinuses residue.



⊥ Maximum/Minimum value

Fig. 2. Changes in the score Penetration-Aspiration Scale of 12 patients at different time points. T0: before treatment, T1: 1 week after treatment, T2: 6 months after treatment.

CSTP, one of the patients was followed up for only 3 years, while the remaining 11 cases were regularly followed up for 5 years. Among the latter, four (33.3%) developed pneumonia, and two were diagnosed with lung metastasis and pneumonitis. The latter two patients later on died. Furthermore, there were two cases of carotid arteriosclerosis and one case of stroke. Relevant comorbidities are listed in Table 4.

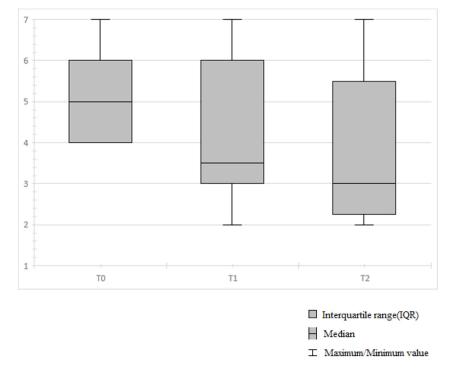


Fig. 3. Changes in the grade of the swallowing performance scale of 12 patients in different time points. T0: before treatment, T1: 1 week after treatment, T2: 6 months after treatment.

	T0 median [IQR]	T1 median [IQR]	T2 median [IQR]	p value		
				T1/T0	T2/T0	T2/T1
SPS	5[4.0-6.0]	3.5[3.0-6.0]	3[2.25-5.5]	.015 <sup>b</sup>	.010 <sup>b</sup>	.059
PAS	4[2.0-7.5]	2[1.0-6.5]	3[1.0-6.0]	.292	.395	.288
Thin liquid 3 mL	32[23.75-39.5]	26[13.0-32.5]	18[5.0-30.0]	.102	.050 <sup>a</sup>	.059
Thin liquid 5 mL	32[26.5-38.0]	31.5[23.5-36.5]	18[12.0-30.0]	.635	$.007^{b}$	.007 <sup>b</sup>
Thick liquid 3 mL	49[34.0-56.0]	44[40.0-48.0]	34[26.0-42.0]	.594	.037 <sup>a</sup>	.017 <sup>b</sup>
Thick liquid 5 mL	49.5[41.75-60.0]	45[38.0-58.0]	39.5[27.0-44.5]	.192	.011 <sup>b</sup>	.027 <sup>a</sup>
Puree 3 mL	38[26.0-48.5]	31[23.0-40.0]	28[14.5-41.0]	.046 <sup>a</sup>	.028 <sup>a</sup>	.345
Puree 5 mL	46[26.0-52.0]	40[20.0-45.0]	19[11.0-48.0]	.176	.075	.400

Table 3. Comparison of swallowing performance at different time points.

Statistical analysis by Wilcoxon's Signed Rank test.

Values are expressed as median [IQR: interquartile range].

T0: before treatment, T1: 1 week after treatment, T2: 6 months after treatment; SPS: Swallowing Performance Scale, FDS: Functional Dysphagia Scale, PAS: Penetration-Aspiration Scale.

<sup>a</sup> Statistical significance: P < 0.05

<sup>b</sup> Statistical significance after Bonferroni correction: P < 0.017.

Table 4. Relevant comorbidities of patients after treatment.

	Т0	T1	T2	T3	
N (%)	12 (100)	12 (100)	12 (100)	12 (100)	
Tube feeding	7 (58.3)	4 (33.3)	3 (25)	4 (36.3)	
Pneumonia	0 (0)	0 (0)	0 (0)	4 (36.3)	
Other oral cancer	0 (0)	0 (0)	0 (0)	2 (16.6)	
Facial cellulitis	0 (0)	1 (8.3)	0 (0)	1 (9)	
Metastasis	0 (0)	0 (0)	0 (0)	2 (18.1)	
Carotid	0 (0)	0 (0)	0 (0)	2 (18.1)	
arteriosclerosis					
Stroke	0 (0)	0 (0)	0 (0)	1 (9)	
Dead	0 (0)	0 (0)	0 (0)	2 (16.6)	
T0: before treatment T1: 1 week after treatment T2: 6					

T0: before treatment, T1: 1 week after treatment, T2: 6 months after treatment, T3: 5 years after treatment.

# 4. Discussion

This study investigated the efficacy of CSTP in the treatment of chronic dysphagia after TC surgery. This treatment program included a combination of 1-h NMES, swallowing muscle strengthening exercises, food texture adjustment, swallowing compensatory posture training, and training to use adaptive feeding devices. Our results revealed that after using this treatment strategy, patients' short- and mid-term swallowing functions improved.

Radiation treatment can induce lingual and pharyngeal muscle fibrosis, diminish laryngohyoid elevation, and reduce opening of the upper esophageal sphincter.<sup>4,5,16</sup> The VFSS before CSTP revealed that more than half of our patients had vallecular residue, pyriform sinus residue and penetration. These findings are consisted with previous reports and indicated that, in addition to difficulty in oral transition following glossectomy, the patients also had pharyngeal dysphagia.<sup>22</sup> Chronic dysphagia in TC patients may be multifactorial, and the above findings may be related to the late effect of radiation, which may cause lingual and pharyngeal muscle fibrosis, and pharyngeal and esophageal stricture. Thus, it reduces the laryngeal elevation and the opening of the upper esophageal sphincter, and causes bolus residue.<sup>23</sup> Sensory receptors in the oropharyngeal region are stimulated by the passage of a bolus and send signals via interneurons to the medullary swallowing center, triggering the swallowing reflex and protecting the airway.<sup>23,24</sup> For patients with chronic feeding tube dependence, a decrease in oral alimentation may further induce disuse atrophy and decrease the sensory feedback of swallowing, thus increasing the risk of aspiration.<sup>24</sup> The increasing the risk of aspiration.<sup>2</sup> associated weakening of the swallowing muscles may increase the difficulty in rebuilding the swallowing function and affect the patient's willingness to participate in exercises.<sup>4,24</sup> Therefor, for patients with chronic dysphagia, swallowing rehabilitation should not only consider increasing muscle strength and improving swallowing performance but also should try to improve the willingness of the participants.<sup>4,23,21</sup>

Normal swallowing muscles include type I and type II muscle fibers, with different distribution ratios depending on the location. A lack of daily swallowing can result in disuse atrophy of the oropharyngeal muscles.<sup>23,24</sup> Type II muscle fibers predominate in high-speed contractions of certain oropharyngeal muscles important for normal swallowing, and may be the first

reflected in disuse atrophy.<sup>4</sup> In addition to muscle atrophy, the central sensory circuit may also be affected in patients with chronic dysphagia.<sup>24</sup> For these reasons, in the CSTP protocol, we considered using NMES therapy alone in the first 30 min. First, NMES stimulates forceful muscle contractions, especially in type II muscle fibers, reverses normal voluntary recruitment, and enhances sensory feedback to the cortical and subcortical swallowing centers, thus promoting swallowing recovery.<sup>15,17</sup> Second, NMES will help achieve passive contraction in patients who are unable to perform oral exercises and movements.

In CSTP, we also combined swallowing muscle strengthening exercises predominantly targeting type I muscle fibers, which can generate greater muscle strength and enhance the therapeutic effect.

In contrast to non-surgical patients, surgery can cause oral defects; thus, compensatory feeding posture and/or adaptive feeding devices were included in the CSTP to overcome the difficulties of food transportation in the oral phase. In CSTP, the posture strategies and/or special adaptive feeding devices were individualized according to the first VFSS. VFSS evaluation of swallowing for patient with chronic dysphagia after glossectomy is necessary for verifying the auto-regulatory mechanisms used by the patient before starting our training program. This study can also help the researchers to define the possible risk of aspiration for their swallowing strategy in real-time swallowing study.<sup>19</sup> Chin-down posture, in addition to improving the basal tongue contact to the posterior pharyngeal wall, was recommended also to increase larvngeal elevation and airway protection.<sup>4</sup> Chin-up and head tilt to the intact side, which provides gravity to assist in bolus transfer to the pharynx, was recommended for partial glossectomies with difficulty in oral transition.<sup>4</sup> For those patients underwent total glossectomy, if their epiglottis movement reduced or completely immobilized, affecting the protection of the airway during swallowing was found in VFSS. Chin - up posture was not recommended, because of increasing risk of aspiration.<sup>26,27</sup> When limitation of mouth opening, increasing oral transit time, limitation of tongue motion, and/ or leakage of bolus content during swallows were observed in VFSS, adaptive feeding

devices including compressible long-mouth bottle, long-arm spoon, syringe and chopsticks were optionally used to improve the efficiency of bolus passing through the oral cavity.<sup>4,19</sup>

Participants in this study were chronic tube-feeding dependence and had ever received different swallowing therapy or home-base swallowing exercise before participating CSTP. They were still unable to eat by mouth. In the evaluation within 1-week after CSTP (T1), the nasogastric tube was successfully removed in three patients (3/7; 42.8%), the improvement may be related to CSTP. For the long-term effect assessment, the nasogastric tube removal rate continued to increase to 57.1% at 6-month follow up after CSTP. Although many restrictions remained on the types of food that could be consumed orally, 58% of patients showed improved SPS after treatment. Even dysphagia was improved in our participants, this study failed to explain for spontaneous recovery because of no control group was included. Further, because of the different treatment regimens patients received, we were unable to use the data from VFSS before CSTP as a comparison of between CSPT and other treatment programs, thus the effectiveness of CSTP for chronic dysphagia in TC patients could not be definitely confirmed.

Our study also demonstrated that the FDS scores of thin and thick liquid, obtained by VFSS analysis were significantly improved compared with those before treatment.

Our positive findings are in consistent with the NMES study for dysphagia in acute phase after HNC treatment.<sup>15</sup> This study demonstrated the limitation of eating different food texture that may be resulted from the damaged of oral structures and mechanical component following surgical treatment.<sup>15</sup> The curative effect of CSTP on chronic dysphagia may be related to the designs of CSTP. In this program, the possible swallowing-related fatigue and the endurance that patients need to complete a meal were taken into the consideration. NMES facilitated the contraction of the disused swallowing muscles in the state of rest. Fatigue during training session can be prevented by shortening synchronize volitional swallowing exercise and cooperated with feeding devices in next stage of treatment, thus compliance can be improved. As their swallowing performance improved, which reflected in the improvement of SPS and FDS, the patients were relatively more willing to cooperate with swallowing exercises. For those whose nasogastric tube could be successfully removed after CSTP, they continued to experience improvements in their results during the 6month follow-up. This may be related with their continued oral feeding practice after the success of removing the feeding tube.

Although the conclusions of studies on the application of NMES to treat dysphagia in patients with HNC remain controversial, most studies have concluded that NMES is effective in the treatment of dysphagia after radiotherapy and chemotherapy and can be regarded as a tool for swallowing training.<sup>13–15</sup> Our results are consistent with those described above, suggesting that NMES can be used as an additional tool to increase the effects of therapy combined with swallowing training for TC with chronic dysphagia. Furthermore, in combination with oropharyngeal exercise, further evaluation of adaptive devices and compensatory techniques is recommended.

The use of swallowing NMES also entails some limitations: it is not suitable for patients with severe neuromuscular damage. Therefore, not all patients with TC can use the CSTP treatment plan and whether the surgical resection site affects the aforementioned muscles and structures must be determined. During the mid-term follow-up period of this study, only four patients (33.3%) had improved PAS after treatment. During the long-term medical record review, four patients developed pneumonia during the 5- year follow-up period after treatment; two of them were mainly due to lung cancer metastasis (which led to lung infection and death). The incidence of pneumonia in this study was 33.3%, which was higher than 23.8–25.4% reported in the literature.<sup>28–30</sup> Dysphagia-related aspiration and aspiration pneumonia are serious and potentially fatal treatment complications in advance HNC patients.<sup>29,30</sup> The significant risk factors of aspiration pneumonia in advance cancer included tube feeding, advance clinical stage, severe dysphagia and incomplete response to treatment.<sup>30</sup> The increased rate of pneumonia may be related to the fact that all the participants in this study had advance cancer with chronic

severe dysphagia. 58.3% of them were nasogastric tube dependence, thus increased risk of aspiration pneumonia.

The main limitation of this study was its small sample size, as this could not enable a more precise analysis of the treatment outcomes with regard to the classification of oral tongue cancer and tongue base cancer or tongue resections of different sizes. In this study, most patients with advanced TC (91.6%), underwent resection of more than half of their tongue were the high-risk group for HNC with chronic dysphagia; consequently, in this small sample size study, most of the patients who participated in the treatment plan may have had better compliance than those who did not meet the inclusion criteria, because we did not compare patients with different compliance, whether there is still a therapeutic effect using this treatment program is undetermined. Additionally, this study did not include a control group with different treatment tools or procedures. Although most of our patients had received swallowing training or instructions before this treatment plan, pre-training was inconsistent and could not be used as a control for this treatment strategy. Therefore, our results may be used as a reference for preliminary reports, and more cases should be included in further research and analysis.

# 5. Conclusion

This study suggested that CSTP has positive effect on improving the swallowing function, based on the SPS and FDS score on the liquid swallowing test in TC patients with chronic dysphagia. CSTP may increase the likelihood of feeding tube removal in TC patient with chronic dysphagia. The results of pre-training VFSS could guide the choice of feeding devices and postural strategies for TC patents with dysphagia.

# **Conflict of interest**

There are no conflicts of interest to declare.

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# References

- 1. National Health Service Cancer Registration Statistics Report 2015-9.
- Ho PS, Ko YC, Yang YHC, Shieh TY, Tsai CC. The incidence of oropharyngeal cancer in Taiwan: an endemic betel quid chewing area. J Oral Pathol Med 2002;31(4):213–9.
- 3. Logemann J, Bytell DE. Swallowing disorders in three types of head and neck surgical patients. *Cancer* 1979;44(3):1095–105.
- 4. Gaziano JE. Evaluation and management of oropharyngeal dysphagia in head and neck cancer. *Cancer Control* 2002 Sep/Oct;9(5):400–9.
- Hutcheson KA, Lewin JS, Barringer DA, Lisec A, Gunn GB, Moore MWS, et al. Late dysphagia after radiotherapy-based treatment of head and neck cancer. *Cancer* 2012;1178:5793–9.
- Kulbersh BD, Rosenthal EL, McGrew BM, Duncan RD, McColloch NL, Carroll WR, et al. Pretreatment, preoperative swallowing exercises may improve dysphagia quality of life. *Laryngoscope* 2006;116:883–6.
- Kotz T, Federman AD, Kao J, Milman L, Packer S, Lopez-Prieto C, et al. Prophylactic swallowing exercises in patients with head and neck cancer undergoing chemoradiation: a randomized trial. Arch Otolaryngol Head Neck Surg 2012;138(4):376–82.
- Zhang LP, Huang ZS, Wu H, Chen WL, Huang ZQ. Effect of swallowing training on dysphagia and depression in postoperative tongue cancer patients. *Eur J Oncol Nurs* 2014;18(6):626-9.
- Nguyen NP, Moltz CC, Frank C, Karlsson U, Smith HJ, Nguyen PD, et al. Severity and duration of chronic dysphagia following treatment for head and neck cancer. *Anticancer Res* 2005;25:2929–34.
- White KT, Easterling C, Roberts N, Wertsch J, Shaker R. Fatigue analysis before and after shaker exercise: physiologic tool for exercise design. *Dysphagia* 2008;23(4):385–91.
- 11. Well M, King E. Patient adherence to swallowing exercise in head and neck cancer. *Curr Opin Otolaryngol Head Neck Surg* 2017;25(3):175-81.
- Brates D, Troche MS, Molfenter SM. The role of fatigue in the aging swallow: a review. OBM Geriat 2021;5(2):1-22.
- Blumenfeld L, Hahn Y, Lepage A, Leonard R, Belafsky PC. Transcutaneous electrical stimulation versus traditional dysphagia therapy: a nonconcurrent cohort study. *Otolaryngol Head Neck Surg* 2006;135:754-7.
- 14. Lin PH, Hsiao TY, Chang YC, Ting LL, Chen WS, Chen SC, et al. Effects of functional electrical stimulation on dysphagia caused by radiation therapy in patients with nasopharyngeal carcinoma. *Support Care Cancer* 2011;9(1):91–9.
- 15. Ryu JS, Kang JY, Park JY, Nam SY, Choi SH, Roh JL, et al. The effect of electrical stimulation therapy on

dysphagia following treatment for head and neck cancer. Oral Oncol 2009;45:665-8.

- Nam HS, Beom J, Oh BM, Han TR. Kinematic effects of hyolaryngeal electrical stimulation therapy on hyoid excursion and laryngeal elevation. *Dysphagia* 2013;28(4):548–56.
- Langmore SE, McCulloch TM, Krisciunas GP, Lazarus CL, Van Daele DJ, Pauloski BR, et al. Efficacy of electrical stimulation and exercise for dysphagia in patients with head and neck cancer: a randomized clinical trial. *Head Neck* 2016;38(S1): E1221–31.
- Wijting Y, Freed M. Training manual for the use of electrical stimulation in the treatment of dysphagia. *Chattanooga group* 2003:103–13.
- Palmer JB, Kuhlemeier KV, Tippett DC, Lynch C. A protocol for the videofluorographic swallowing study. *Dysphagia* 1993;8:209–14.
- Rosenbek JC, Robbins JA, Roecker EB, Coyle JL, Wood JL. A penetration-aspiration scale. *Dysphagia* 1996;11:93-8.
- Han TR, Park NJ, Park JW. Quantifying swallowing function after stroke: a functional dysphagia scale based on videofluoroscopic studies. *Arch Phys Med Rehabil* 2001;82(5):677–82.
- 22. Stenson KM, MacCracken E, List M, Haraf DJ, Brockstein B, Weichselbaum R, et al. Swallowing function in patients with head and neck cancer prior to treatment. Arch Otolaryngol Head Neck Surg 2000;126(3):371–7.
- 23. King SN, Dunlap NE, Tennant PA, Pitts T. Pathophysiology of radiation-induced dysphagia in head and neck cancer. *Dysphagia* 2016;31:339–51.
- Mehdizadeh OB, Dhar SI, Evangelista L, Nativ-Zeltzer N, Bewley AF, Belafsky PC, et al. Prevalence of profound laryngeal sensory neuropathy in head and neck cancer survivors with feeding tubedependent oropharyngeal dysphagia. *Head Neck* 2020;42(5):898–904.
- 25. Wu YS, Lin PY, Chien CY, Fang FM, Chiu NM, Hung CF, et al. Anxiety and depression in patients with head and neck cancer: 6-month follow-up study. *Neuropsychiatric Dis Treat* 2016;12:1029–36.
- Calvo I, Kirstyn L, Sunday BS, Humbert IA. Effect of chin-up posture on the sequence of swallowing events. *Head Neck* 2017;39:947–59.
- events. *Head Neck* 2017;39:947–59.
  27. Halczy-Kowalik L, Wiktor A, Rzewus A, Kowalczyk R, Wysocki R, Posio V. Compensatory mechanisms in patients after a partial or total glossectomy due to oral cancer. *Dysphagia* 2015;30: 738–50.
- Xu B, Boero IJ, Hwang L, Le QT, Moiseenko V, Sanghvi PR, et al. Aspiration pneumonia after concurrent chemoradiotherapy for head and neck cancer. *Cancer* 2015 Apr;121(8):1303–11.
- Mortensen HR, Jensen K, Grau C. Aspiration pneumonia in patients treated with radiotherapy for head and neck cancer. *Acta Oncol* 2013;52: 270-6.
- 30. Shirasu H, Yokota T, Hamauchi S, Onozawa Y, Ogawa H, Onoe J, et al. Risk factors for aspiration pneumonia during concurrent chemoradiotherapy for head and neck cancer. *BMC Cancer* 2020;20(1): 1–10.