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CASE REPORT

Esophageal Speech for a Patient with Amyotrophic Lateral Sclerosis Who Underwent a Central-part Laryngectomy to Prevent Aspiration: A Case Report

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Background: To prevent aspiration, patients with irreversible dysphagia may undergo surgeries that separate the esophagus and trachea. Such interventions result in loss of vocal function and require alternative communication methods. We report a patient with amyotrophic lateral sclerosis (ALS) who used esophageal speech after receiving a central-part laryngectomy (CPL) to prevent aspiration. **Case:** A 64-year-old woman with ALS was admitted to our hospital. The patient maintained good cognitive and oral function and presented with mild dysarthria and dysphagia. Faced with rapidly worsening respiratory distress, saliva aspiration, and excessive sputum, she underwent a tracheostomy on the premise of invasive ventilation. Subsequently, the patient began using a voice-generating application for communication. Given the patient's sincere hope to prevent aspiration and aspiration pneumonia, achieve safe oral intake, and decrease caregiver burden for frequent suctioning, the patient underwent a CPL. Following surgery, belching was observed during meals, and the patient could phonate when she belched. This finding led to four speech therapy sessions to practice esophageal speech, allowing the patient to use the pseudo-speech technique for short conversations. Removal of the entire cricoid cartilage in the CPL decreases the upper esophageal sphincter (UES) pressure, thereby allowing air to easily pass through the UES. Therefore, the patient could use the air as a sound source for esophageal speech without extensive training. **Discussion:** Esophageal speech may be an alternative to oral communication in patients undergoing CPL. Further research is warranted to generalize these findings to patients undergoing CPL.

Key Words: alternative augmentative communication; amyotrophic lateral sclerosis; case report; central-part laryngectomy; esophageal speech

INTRODUCTION

Amyotrophic lateral sclerosis (ALS) is a fatal neurodegenerative disease that affects upper and lower motor neurons, resulting in progressive weakness and atrophy of limb and respiratory muscles.[1] ALS also damages swallowing and speech-related muscles, causing dysphagia and dysarthria during disease progression. Dysphagia or difficulty swallowing leads to serious health consequences such as malnutrition,[2] aspiration pneumonia,[3] or death[4] in patients with ALS. Furthermore, the loss of safe eating and effective communication significantly diminishes the quality of life of patients.[5,6]

Surgical treatment is an option for individuals with severe irreversible dysphagia, including those with ALS, to prevent recurrent aspiration and airway complications. Total laryngectomy, laryngotracheal diversion, and tracheoesophageal anastomosis are standard surgical procedures.[7,8] By separating the trachea and esophagus, these surgeries successfully

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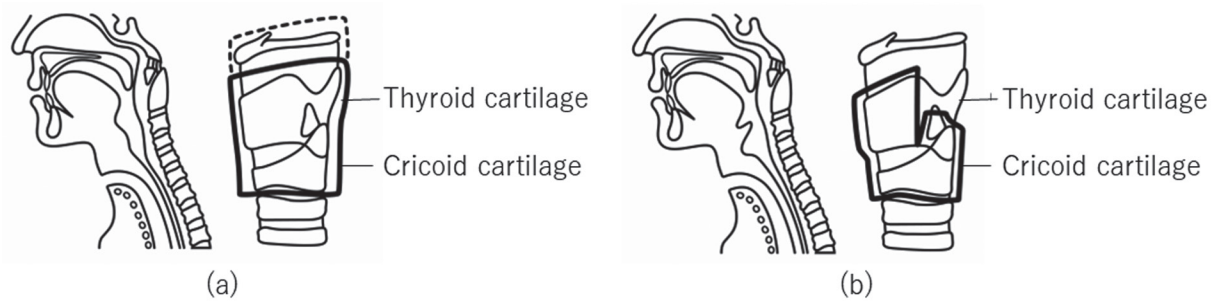

Fig. 1. Total laryngectomy (a) and central-part laryngectomy (b). The bold lines encircle the removed areas. The dashed line represents the possible removal area.

manage life-threatening aspirations; however, these methods also result in loss of vocal function. Less invasive methods, such as central-part laryngectomy (CPL) or glottic closure, are preferable for patients with severe dysphagia and advanced respiratory impairment.[7,9–11] A CPL removes the middle part of the thyroid cartilage and the entire cricoid cartilage while preserving the lateral aspect of the thyroid cartilage and epiglottis[10] (**Fig. 1**). Removing the whole cricoid cartilage leads to decreased upper esophageal sphincter (UES) resting pressure and prolongs the relaxation duration of UES.[10]

Given the effects of aspiration-prevention surgeries, patients need to use augmentative and alternative communication (AAC) strategies. Multiple AAC tools are available after the loss of vocal function, including pen and paper, photos/pictures/symbols/word and alphabet boards, touch-screen devices, mobile applications (apps) on smartphones, and speech-generating devices on a tablet or computer.[12]

To restore oral communication, an electrolarynx is an option for alaryngeal speech.[13] This small hand-held vibrating device is placed against the neck or cheek, and the vibration that is introduced into the oral cavity is used as a sound source for speech. These tools are simple to operate and do not require extensive training. However, patients with ALS have difficulty handling these tools because of limited motor control of the upper limbs and oral function.[14] Therefore, more complex AAC devices, such as eye-tracking systems, are required for communication.[15]

Esophageal speech is another technique of alaryngeal speech. Patients who have undergone laryngectomy for cancer treatment produce speech sounds with airflow-induced vibrations of the pharyngoesophageal segment (**Fig. 2**). This simple method does not require external battery-powered devices or hands for operation and is widely used by laryngectomees after laryngeal cancer.[16,17] However, there are no reports on esophageal speech in patients undergoing aspiration-prevention surgery. The hands-free capability of esophageal speech may be a valuable option for patients with limited limb function who lose voice after surgery.

This single case report describes the utility of esophageal speech in a patient with ALS who lost her voice after receiving CPL to prevent aspiration. This report details the benefits and precautions of utilizing esophageal speech after CPL.

CASE

Patient Information

This case study followed CARE guidelines[18] for patient reporting. Written informed consent was obtained from the patient.

A 64-year-old female patient was diagnosed with ALS and admitted for a gastrostomy in August 2021. Before hospital admission, the patient was ambulatory and had lived with her husband at home. She received noninvasive positive-pressure ventilation support during the night. She was on a regular diet and took thin liquids with occasional cough.

Clinical Findings and Diagnostic Assessment

Soon after admission, the patient was put on nil per os because of rapidly worsening respiratory distress and excessive sputum. She was also placed on bed rest because of suspected ampulla cardiomyopathy. On day 3 of hospitalization, the patient was referred to a rehabilitation center. The patient maintained good cognitive function (Mini-Mental State Examination[19]: 30/30 points) and communicated verbally with an overall intelligibility grade of 1 on a 5-point Likert scale (1, excellent; 5, not intelligible).[20] Mild nasality was observed during her speech. Her limb muscle strength was significantly reduced (manual muscle testing[21]: upper extremity, grade 2; lower extremity, grade 3). Her limited muscle strength and

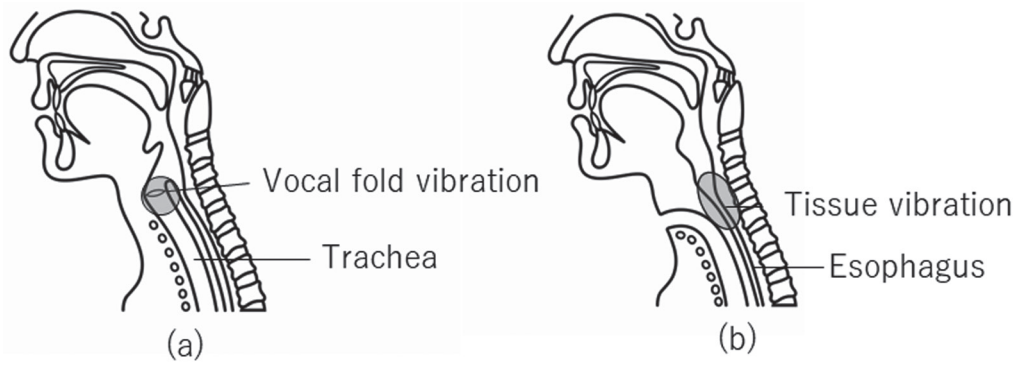

Fig. 2. Normal speech production (a) and esophageal speech production (b). The gray areas encircle the sound source for speech production.

dexterous hand movements required considerable assistance with daily living (Barthel Index[22]:10/100 points).

A physiotherapist (PT) and occupational therapist (OT) initiated rehabilitative interventions to maintain the patient's physical condition and daily activities. In addition, the OT set up voice-generating apps on the patient's smartphone and a switch-controlled device on a tablet for future alternative communication.

The gastrostomy was postponed because the patient's respiratory condition worsened after hospitalization. The excess sputum required frequent intratracheal suction for airway safety. On day 18, the patient underwent a tracheostomy to provide access for intratracheal suction and invasive ventilation, resulting in temporal loss of voice. The patient continued to communicate via lip-reading and AAC devices.

A gastrostomy was performed on day 38. However, persistent cough when swallowing saliva became evident, requiring more frequent suctioning. To be free from the anxiety of aspiration and aspiration pneumonia, ensure safe oral intake, and decrease caregiver burden for frequent suctioning, the patient indicated her desire to undergo CPL on day 47.

On day 60 [postoperative day (POD) 13], the attending otolaryngologists implemented videofluoroscopy and identified mild dysfunction in laryngeal elevation, pharyngeal squeeze, and UES opening. In test swallows of 5 mL of moderately thick and thin test bolus, no leakage, penetration, aspiration, or nasal regurgitation was observed. A mild pharyngeal residue was noticed, which was cleared by multiple swallows. Postoperative high-resolution manometry on day 60 revealed decreased UES resting pressure compared to the preoperative examination (**Fig. 3**). In addition, an increased UES pressure at the initiation of swallowing was observed in the preoperative examination but diminished after surgery.

Therapeutic Interventions for Restoring Speech after CPL

Figure 4 displays a timeline of rehabilitative interventions that were used during hospitalization to restore the voice of the patient. A speech therapist (ST) started rehabilitative intervention on day 61 (POD 14). The patient demonstrated a good range of motion in the lips and tongue. The patient was fed via a gastrostomy tube and restarted oral intake with jelly-like food. The diet was gradually upgraded to a regular diet by day 88 (POD 41). During meals, frequent belching was observed, causing discomfort to the patient. Nurses degassed via the gastrostomy tube after each meal.

The patient continued communicating with the medical staff using lip-reading and AAC devices. However, the patient's husband could not read the patient's lip movements. The ST then introduced an electrolarynx, which produced good intelligible speech when placed on the patient's cheek (grade 2 on a 5-point Likert scale: sometimes unintelligible). The husband considered the electrolarynx helpful as a communication tool. However, the patient did not want to use the device because she needed to ask her husband to place the electrolarynx on her cheek when she wanted to speak.

On day 80 (POD 33), the PT noticed the patient speaking short phrases upon belching when drinking coffee. This observation led the ST to arrange a trial training session for esophageal speech as a hands-free and device-free form of verbal communication. Before the session, the patient's attending physiatrist and otolaryngologist confirmed that no contraindications had been observed in the previous laryngoscopy or videofluoroscopy, such as spasms on the pharyngoesophageal segment.

On day 85 (POD 38), the ST instructed the patient to close her lips tightly to increase air pressure in the oral cavity,

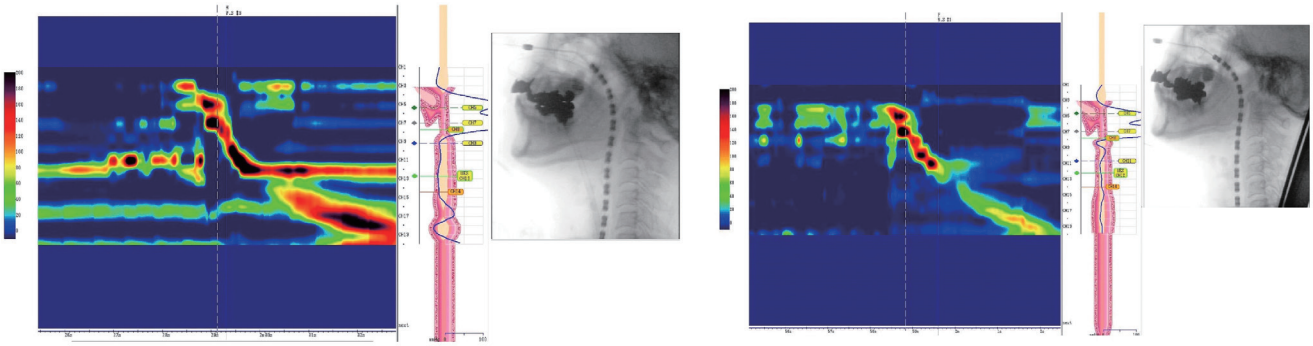

Fig. 3. High-resolution manometry: pre-operation (a) and post-operation (postoperative day 13) (b). Postoperative high-resolution manometry (b) revealed that UES resting pressure was low. The UES pressure at the initiation of swallowing was high in the preoperative examination (a) but diminished after surgery (b).

Hospital days		1 5 10 15 20 25 30 35 40 45 50 55 60 65 70 75 80 85 90
Surgeries		tracheostomy / PEG / CPL
Rehabilitation		PT/OT / ST
Communication	speech	voice / electro larynx, esophageal speech
	device	writing, a voice-generating mobile app, touch-screen device

Fig. 4. Timeline of rehabilitative interventions and alternative methods of communication. PEG, percutaneous endoscopic gastrostomy.

pump the air to the esophagus (i.e., the injection method), and open the mouth and belch. She quickly mastered this technique because she had already encountered belching as a complication of aspiration-prevention surgery. The patient then practiced producing single vowels. The practice was upgraded to two-syllable words such as /ao/ (blue), /mame/ (beans), and several selected words such as /arigato/ (thank you), /onegai/ (please), and /kyuin/ (suctioning) for short daily conversations. At the end of four training sessions, speech intelligibility was graded as 2 (sometimes unintelligible) on a 5-point Likert scale. The patient was encouraged to use esophageal speech when talking with the medical staff and her husband. On day 92 (POD 45), the patient was transferred to a hospital to arrange social services needed to return home.

Patient Perspective

The patient reported that esophageal speech was helpful in short conversations with caregivers to ask for assistance and express thanks without relying on others to set up a communication tool, unlike an electrolarynx. She valued voice-producing apps and touch-screen devices as methods for extended conversations. The patient did not report excessive effort or fatigue in producing a pseudo-voice.

DISCUSSION

Here, we first report the case of a patient with ALS who used esophageal speech for daily communication after a CPL was performed to prevent aspiration. The patient found esophageal speech to be convenient because it did not require the use of hands or a device. This finding is unique because it suggests that esophageal speech, commonly performed by laryngectomy patients for hypopharyngeal or laryngeal cancer, can also be used by patients after undergoing surgery to prevent intractable aspiration caused by neurological disease.

To our knowledge, there has been no report on the utility

of esophageal speech after aspiration-prevention surgery, including CPL. Several conditions exist for the successful use of esophageal speech after laryngectomy to prevent aspiration. First, oral function should be maintained for good speech intelligibility. Unlike laryngectomy patients with laryngeal cancer, patients undergoing aspiration-prevention surgeries are likely to present with dysarthria because of motor dysfunction of speech-related muscles. Even if the voice source is substituted with pharyngoesophageal tissue vibration, the patient's speech may be dysarthric and poorly intelligible. Before surgery, our patient demonstrated good tongue, lip, and alveolar movements. Therefore, after surgery, well-maintained oral functions resulted in clear speech sounds once voice production was restored. However, it should be noted that the patient's speech intelligibility will deteriorate with disease progression because of the deterioration of muscles for speech production.

Second, belching, a disturbing postoperative complication of laryngectomy, can be advantageous for mastering esophageal speech. Belching is the audible escape of air from the esophagus or stomach into the pharynx,[23] affecting more than 50% of patients who undergo laryngectomy[24,25] including CPL.[26] Removing the whole cricoid cartilage in total laryngectomy or CPL facilitates UES relaxation and decreases UES pressure, leading to an influx of air to the esophagus or stomach (aerophagia).[10] Esophageal dysmotility may further contribute to the retention of air in the esophagus. Air retained in the upper gastrointestinal tract should escape to the pharynx to avoid an accumulation of air remaining in the stomach.

A postoperative high-resolution manometry study of our patient revealed that the resting UES pressure was low. No increase in UES pressure at the initiation of swallowing was detected. These postoperative changes may have resulted in the influx of air during meals[26] to the esophagus and stomach, allowing the patient to use the air as a sound source for esophageal speech without extensive training. Therefore, patients who undergo total laryngectomy or CPL and have esophageal dysmotility may learn esophageal speech with minimal time and effort[27] because injecting air into the esophagus is the first and most challenging step in esophageal speech.

This case demonstrated several benefits of esophageal speech as an alternative communication method after laryngectomy, specifically for those with difficulty handling communication devices because of upper limb dysfunction. Given that esophageal speech does not require a device to be set up, the method enables timely communication without depending on the assistance of others. Esophageal speech also allows the patient's communication partners to feel that the patient "speaks" to them rather than a voice-generating device. However, esophageal speech has several limitations. The volume of air that can be injected at one time is low (70–100 mL), which means that only a few syllables can be produced simultaneously with limited voice volume. However, even this reduced voice volume and length enables short conversations with caregivers. Multiple AAC strategies should be used for more elaborate communication. The choice of other AAC methods should be tailored individually, considering physical, cognitive, and oral functions, prognosis, and patient and family expectations.

This study had several limitations. Because this was a clinical case report, some variables could not be controlled. For example, we provided only four sessions of esophageal speech training, and long-term follow-up was not possible after the patient was transferred to another hospital. In addition, observer bias was inevitable because the investigators were involved in the patient's treatment. For example, an ST and physiatrist who treated the patient daily assessed speech intelligibility. However, the two assessors scored the patient's speech using clear definitions for evaluating the patient's speech to minimize bias. Despite these limitations, this case report contributes to the literature by demonstrating the utility of esophageal speech after CPL.

Further research is warranted to investigate the utility of esophageal speech in a longitudinal study with long-term follow-up. For generalizability, the results should be reported in a case series. Along with functional outcomes, assessing patients' and caregivers' perceptions of communication and quality of life would be beneficial.

CONCLUSION

Esophageal speech may be an alternative communication method for patients undergoing CPL to prevent aspiration. The preliminary findings of this case report will guide future research to examine the generalizability of esophageal speech after this surgical procedure.

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CONFLICTS OF INTEREST

The authors declare no conflicts of interest.

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