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CASE REPORT

A Case of Dysphagia and Dysarthria Improved by Flexible-palatal Lift/augmentation Combination Prosthesis

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Background: Palatal augmentation prosthesis (PAP) and palatal lift prosthesis (PLP) have been used to improve dysphagia and dysarthria. However, to date, there are few reports on their combined use. We report a quantitative evaluation of the effectiveness of a flexible-palatal lift/augmentation combination prosthesis (fPL/ACP) based on videofluoroscopic swallowing study (VFSS) and speech intelligibility testing. **Case:** An 83-year-old woman was admitted to our hospital with a hip fracture. She developed aspiration pneumonia at 1 month after partial hip replacement. Oral motor function tests revealed a motor deficit of the tongue and soft palate. VFSS showed delayed oral transit, nasopharyngeal reflux, and excessive pharyngeal residue. The cause of her dysphagia was assumed to be pre-existing diffuse large B-cell lymphoma and sarcopenia. To improve the dysphagia, an fPL/ACP was fabricated and applied. It improved the patient's oral and pharyngeal swallowing and speech intelligibility. In addition to prosthetic treatment, rehabilitation and nutritional support allowed her to be discharged. **Discussion:** The effects of fPL/ACP in the present case were similar to those of flexible-PLP and PAP. f-PLP assists in elevation of the soft palate and improved the nasopharyngeal reflux and hypernasal speech. PAP promotes tongue movement and results in improved oral transit and speech intelligibility. Therefore, fPL/ACP may be effective in patients with motor deficits in both the tongue and soft palate. To maximize the effect of the intraoral prosthesis, a transdisciplinary approach with concurrent swallowing rehabilitation, nutritional support, and physical and occupational therapy is necessary.

Key Words: dysarthria; dysphagia; palatal augmentation prosthesis; palatal lift prosthesis; videofluoroscopic swallowing study

INTRODUCTION

The palatal augmentation prosthesis (PAP) and palatal lift prosthesis (PLP) are prosthetic modalities used to compensate for oropharyngeal motor deficits in dysphagia and dysarthria. PAP has been developed as a treatment for tissue defects that develop after oral cancer surgery and is often applied in patients with neurogenic motor deficits caused by diseases such as amyotrophic lateral sclerosis or stroke.[1–3] It shortens the bolus transit time, reduces pharyngeal residue, and improves speech intelligibility. PLP resolves velopharyngeal insufficiency caused by soft palate defects and motor deficits and improves hypernasal speech.[4] These devices are generally fabricated independently because each plays a different role. However, two cases of a combined palatal lift and augmentation prosthesis have been reported. In one case, the

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combined prosthesis was used to improve dysarthria after stroke.[5] In the second case, for which it was used to manage dysphagia and dysarthria after surgery for chondrosarcoma,[6] although the improvement of dysarthria was emphasized, improved bolus transit was the only improvement observed for swallowing function. Currently, little is known regarding the effects of integrated PAP and PLP devices on swallowing function. One reason for this may be the inhibition of swallowing movements by the hard lift of PLP. Forced elevation of the soft palate by a palatal lift causes severe discomfort during swallowing.[7] Ohno et al. fabricated a flexible-lift PLP (f-PLP) to overcome this issue.[8] The effects of the f-PLP on swallowing function include the elimination of nasopharyngeal reflux and increased velopharyngeal pressure, as demonstrated through high-resolution manometry.[8,9]

Here, we describe the case of a patient with dysphagia and dysarthria accompanied by motor deficits of the tongue and soft palate. A flexible-palatal lift/augmentation combination prosthesis (fPL/ACP), which is a combination of the f-PLP and PAP, was used to improve her dysphagia and dysarthria. We report a quantitative evaluation of the effectiveness of the fPL/ACP through videofluoroscopic swallowing study (VFSS) and testing of speech intelligibility.

CASE

An 83-year-old woman was admitted to Seirei Mikatahara General Hospital with a hip fracture on the left side. She was quite thin before admission, with a height of 157.6 cm and a weight of 33.0 kg (body mass index [BMI], 13.3 kg/m^2). She lived alone and independently performed her activities of daily living. The patient had a history of diffuse large B-cell lymphoma (DLBCL) for 6 years. DLBCL had been treated with chemotherapy, and relapse and remission had occurred three times. The patient had experienced oral problems, such as numbness of the mouth and dysgeusia, since the onset of DLBCL, but she had no history of aspiration pneumonia.

Figure 1 presents the course of treatment. The patient received partial hip replacement on day 7, and physical and occupational therapy was initiated. She received a regular diet in the upright position. The patient developed aspiration pneumonia on day 53. The blood test showed that the

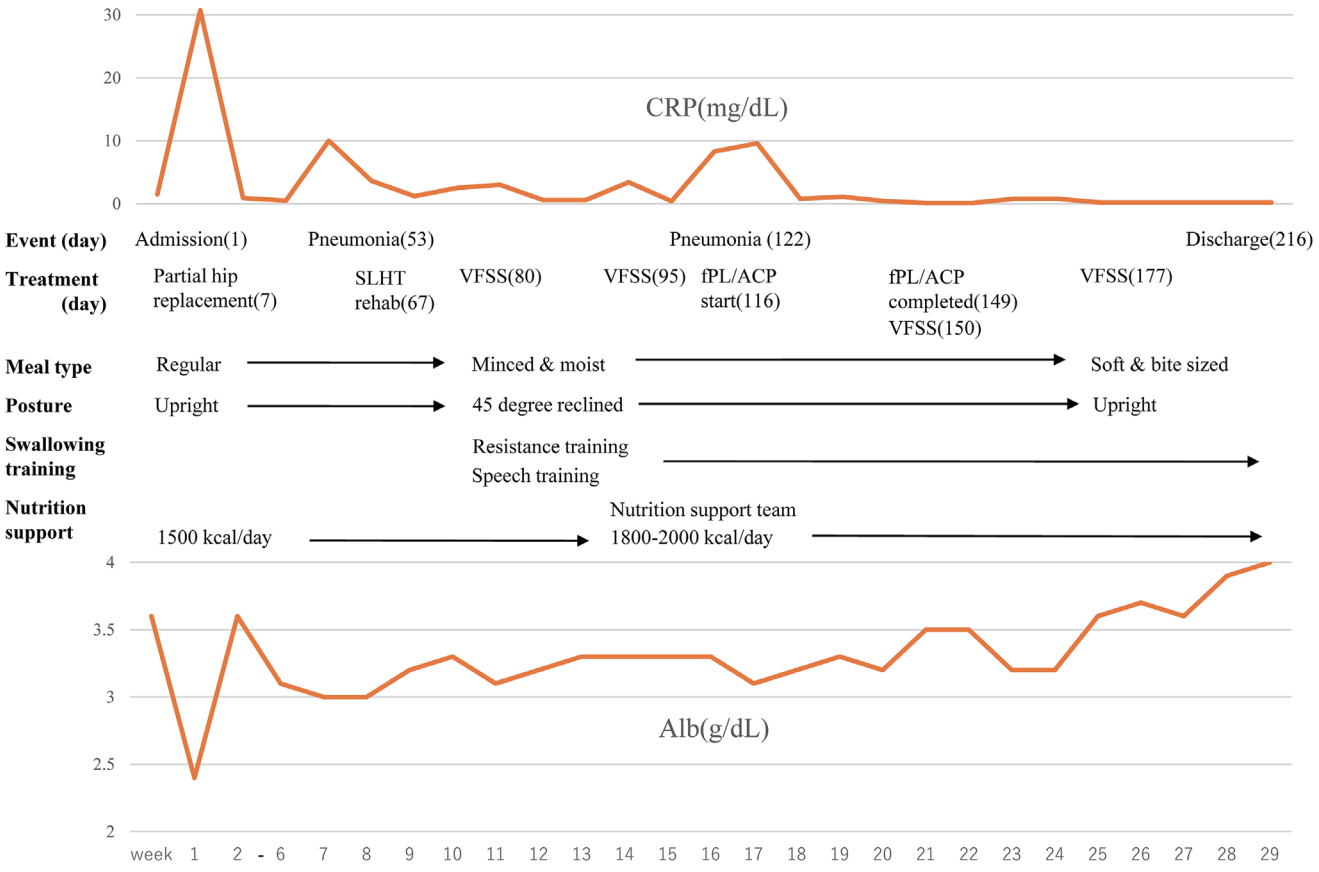

Fig. 1. Course of patient treatment.

C-reactive protein (CRP) levels were elevated to 10.0 mg/dL, and a chest radiograph revealed an opacity in the left lower lobe. Subsequently, antibiotic treatment was initiated, and the CRP levels were improved.

A speech language hearing therapist (SLHT) initiated evaluation of the patient's swallowing and speech function from day 67. She had no consciousness disorder and scored 15 points on the Glasgow coma scale. She had a score of 24 points on the Mini-Mental State Examination and had a point reduction on a calculation task. The patient had a limited range of motion in her left hip because of a fracture. There was no obvious motor paralysis or ataxia in the four limbs. Although the patient had numbness in both plantar facia prior to hospitalization, no other sensory deficits were apparent in the limbs or trunk. The patient had a regular diet in the upright position but presented with a wet voice and coughing after swallowing. The range of tongue movement was almost maintained, but the movement was slow. The elevation of the soft palate was insufficient with no laterality. An oral diadochokinesis of the /ka/ sound was of 3.6 times/s, which was lower than the healthy average of 6.0 times/s.[10] The maximum phonation time (MPT) was 10.7 s with a hypernasal speech. Her speech was partially comprehensible and often required speculation by the listener. Touching the soft palate, uvula, and tongue base with a tongue depressor did not induce gag reflex. As far as we could assess, there was no obvious cranial nerve disorder other than dysarthria and dysphagia.

Fiberoptic endoscopic evaluation of swallowing was performed on day 71. Insufficient elevation of the soft palate and nasopharyngeal reflux of food was observed. There was no laterality in pharyngeal contraction. There was no vocal cord paralysis or saliva aspiration. A large amount of thickened water remained in the pharynx and was not removed by subsequent swallowing. There was no aspiration of food. A VFSS on days 80 and 95 showed delayed bolus transit and oral residue in the oral phase. During the pharyngeal phase, nasopharyngeal reflux of the bolus was observed. The pharyngeal contraction was weak. This patient was unable to transport the bolus to the hypopharynx because of impaired bolus transfer with the tongue, nasopharyngeal reflux, and weakened pharyngeal contractions. The bolus was passed back and forth between the vallecula and the oral cavity with each swallow, resulting in a large amount of vallecular residue. Both thick and thin water showed laryngeal penetration, with a penetration aspiration scale (PAS) score of 2 points.[11] The patient had a BMI of 12.8 kg/m^2 and a grip strength of 9.5 kg. The patient's muscle mass was 4.5 kg/m^2 as measured by bioelectrical impedance analysis (BIA; InBody S-10, InBody, Tokyo, Japan). A blood test on day 80 showed a low serum albumin (Alb) level of 3.0 g/dL. The BMI, grip strength, and muscle mass values met the diagnostic criteria for sarcopenia provided by the Asian Working Group for Sarcopenia.[12] Tongue pressure was measured with a tongue pressure measurement device (TPM-01, JMS, Hiroshima, Japan) and was low at 7.5 kPa, indicating a decreased tongue muscle strength. The definite cause of her dysphagia was unclear.

Because a long time had passed since the onset of DLBCL and the patient was able to eat a regular diet independently prior to admission, it was assumed that the dysphagia surfaced postoperatively for some reason. Given the surgical invasion, decreased activity, low BMI values, low grip strength, and loss of muscle mass, we hypothesized that sarcopenia may have worsened during hospitalization. Based on these findings, the swallowing rehabilitation team suspected that the dysphagia was caused by pre-existing DLBCL and sarcopenia. The patient's food was modified to a minced and moist texture. In addition, swallowing and speech training and increased nutritional intake were initiated by the SLHT and nutrition support team, respectively. The nutritional care plan was reviewed by a registered dietitian: the patient was offered a dysphagia diet with 1500 kcal/day and either ice cream or high calorie jelly as an add-on food with each meal. These had a total nutritional content of 500 kcal/day. The patient was able to eat without leaving any food behind. The rehabilitation physician, dentist, and SLHT discussed the case and decided to fabricate an fPL/ACP as a symptomatic treatment for the patient's oropharyngeal problems.

The dentist fabricated the fPL/ACP between days 116 and 149. The fPL/ACP comprised an augmented palate, four wire clasps, and a silicone flexible palatal lift (**Figs. 2 and 3**).[8] The fPL/ACP is a prosthesis that combines a soft palatal lift behind an augmented palatal plate. Given that the patient did not own a denture, the dentist fabricated a new resin palate and modified its thickness using denture lining material (Tissue Conditioner II; SHOFU, Kyoto, Japan) to assist tongue movement. The posterior end of the lift was set at the base of the uvula. The lift was raised to a height near the palatal plane. The contact between the posterior pharyngeal wall and the lift was checked using an endoscope and a nasal emission, and the width and elevation angle of the lift were adjusted. A palatal lift was applied to the resin palate to adjust the elevation of the soft palate while avoiding pain and discomfort. Finally, the denture liner material was replaced with resin, and the lift was replaced with a silicone flexible

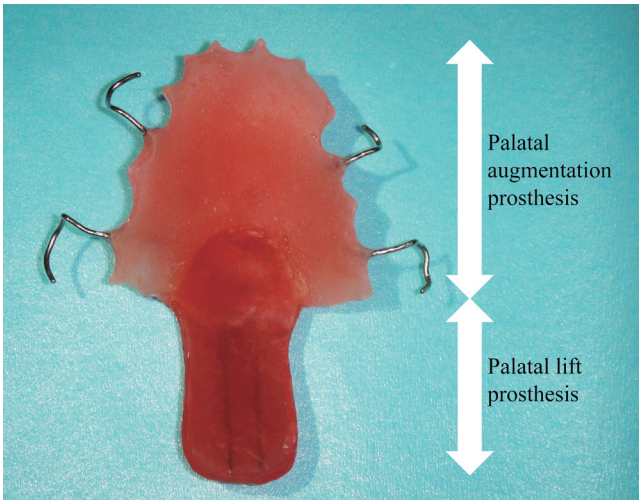

Fig. 2. Flexible-palatal lift/augmentation combination prosthesis prepared for our patient.

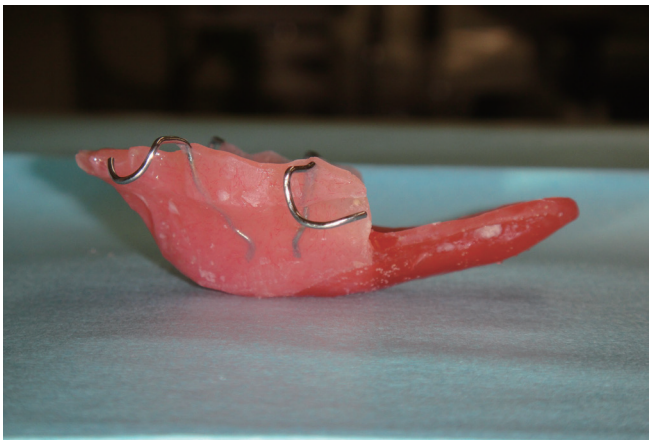

Fig. 3. Lateral view of the flexible-palatal lift/augmentation combination prosthesis.

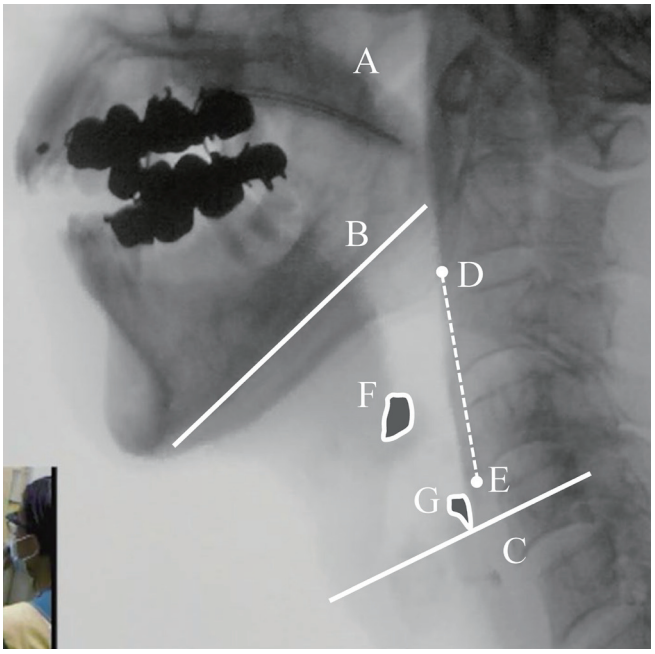

Fig. 4. Fluoroscopic image showing reference points used for measurements during videofluoroscopic swallowing study. A, soft palate; B, point where the lower edge of the mandible crosses the tongue base; C, upper esophageal sphincter; D, anterior inferior corner of C2 vertebra; E, anterior inferior corner of C4 vertebra; F, vallecular residue; G, pyriform sinus residue.

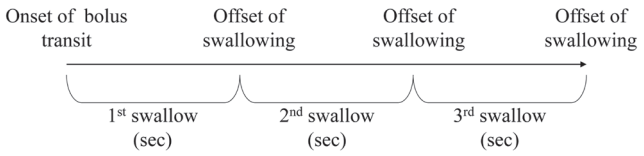

Fig. 5. Timeline showing the measurement of "swallowing time."

lift. To assess the effects of the fPL/ACP, the following evaluations were performed.

Dysphagia

VFSS findings on day 177 and tongue pressure were compared with and without the fPL/ACP. The dentist was present at the VFSS to assess the effectiveness of the prosthesis and the need for adjustment. Pharyngeal residue, oral transit time (OTT), pharyngeal transit time (PTT), swallowing time, and nasopharyngeal reflux were measured.[13] **Figures 4 and 5** present the method used during VFSS. Baribright P powder (Kaigen Pharma, Osaka, Japan) was used for the barium test. In brief, 350 g of Baribright P powder was adjusted to 150 w/v% with 145 mL of water. For the VFSS test food, 240 mL of water and thickening agent (Neo-High Toromeal III, Foodcare, Kanagawa, Japan) was added to 160 mL of barium solution. The viscosity of the test food was adjusted to 300–500 mPa·s according to the recommendations of the Japanese Society of Swallowing Rehabilitation. The patient was administered 3 mL of the test food using a teaspoon and asked to swallow it. She was asked to swallow again if she felt a subjective residual sensation on the initial swallow. The OTT indicates the duration from the beginning of bolus transport on the tongue until the tail of the bolus passed over the intersection of the lower end of the mandible and the tongue base. The PTT indicates the duration from when the tip of the bolus passed over the intersection of the lower end

Table 1. Comparison of VFSS findings with and without fPL/ACP on day 177

Number of swallows	NRRS v	NRRS ps	OTT (s)	PTT (s)	Swallowing time (s)	Nasopharyngeal reflux
Without fPL/ACP						
First	1.25	0.00	3.3	2.6	4.0	Present
Second	0.76	0.00			9.1	Present
Third	0.42	0.00			15.1	Present
With fPL/ACP						
First	0.48	0.00	0.7	0.6	1.7	Absent
Second	0.15	0.34			6.6	Absent
Third	0.25	0.00			10.9	Absent

v, vallecular; ps, pyriform sinus.

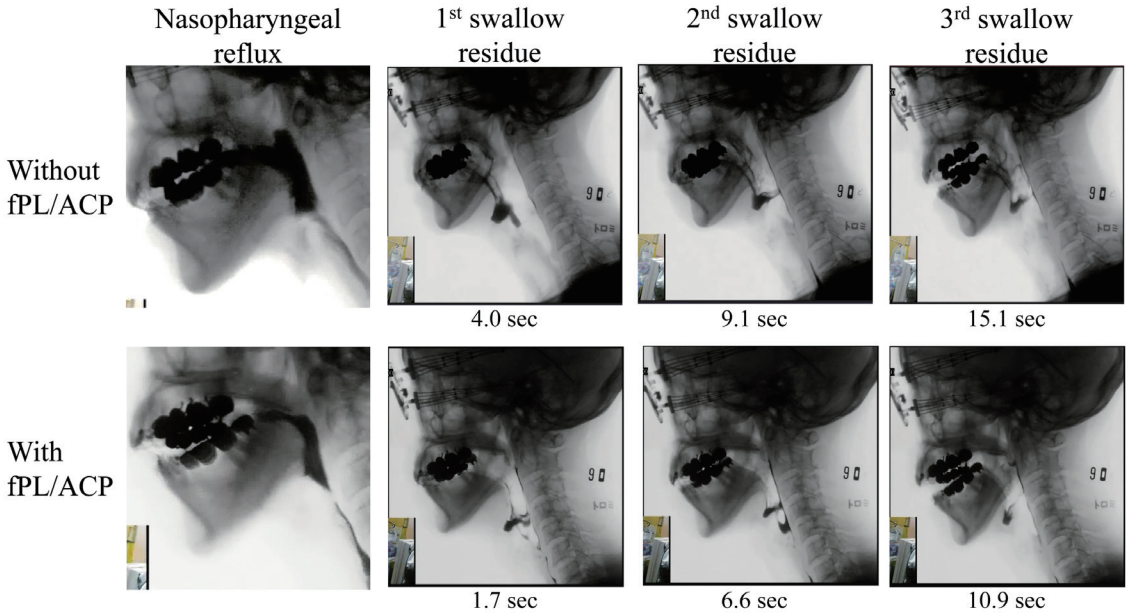

Fig. 6. Comparison of nasopharyngeal reflux and bolus residue by VFSS. Top: without fPL/ACP; bottom: with fPL/ACP.

of the mandible and the tongue base until the tail of the bolus passed over the upper esophageal sphincter (UES). The swallowing time was the time from the beginning of bolus transport until the laryngeal elevation caused by swallowing returned to the resting position; if subsequent swallowing occurred, the time from the end of the previous swallow to the end of the next swallow was measured. Pharyngeal residue in the vallecula and pyriform sinus was assessed using the Normalized Residue Ratio Scale (NRRS).[14] The NRRS is based on a line connecting the anterior inferior corner of the C2 vertebra and the anterior inferior corner of the C4 vertebra. The amount of residue occupying a square with the reference line as one side was measured. Nasopharyngeal reflux was evaluated as present or absent. The maximum tongue pressure was the average of three measurements.

Dysarthria

A speech intelligibility test was performed on day 154.[15] The speech sample was a recording of the patient's speech of 100 Japanese phonemes and 100 words. Four SLHTs listened to the recordings, and the speech intelligibility was defined as the percentage of correctly perceived speech. The MPT was measured for a sustained /a:/ duration.

The effects of the fPL/ACP on swallowing function are shown in **Table 1** and **Fig. 6.** The fPL/ACP reduced vallecular residue as measured by NRRS for each swallow, including the subsequent swallows (without fPL/ACP: 1.25–0.76–0.42, with fPL/ACP: 0.48–0.15–0.25). The fPL/ACP improved bo-

Table 2. Effect of fPL/ACP on phonation and articulation on day 154

	Without fPL/ACP	With fPL/ACP
Speech intelligibility		
100 Japanese phonemes (%)	68.6 ± 10.8	73.3 ± 2.3
Bilabial consonants (%)	68.0 ± 9.8	83.0 ± 7.7
Denti-alveolar consonants (%)	73.3 ± 12.9	77.0 ± 3.3
Velar consonants (%)	25.0 ± 16.8	25.0 ± 16.2
100 words (%)	95.5 ± 3.6	98.5 ± 0.9
Maximum phonation time (s)	10.7	12.8

Data for intelligibility are given as mean ± standard deviation (n = 4).

lus transport and increased the amount of bolus reaching the area near the UES. Considering the temporal measurements, the OTT (without/with: 3.3/0.7 s) and PTT (without/with: 2.6/0.6 s) were shortened on wearing the fPL/ACP, as was the swallowing time for three swallows (without fPL/ACP: 4.0–9.1–15.1 s, with fPL/ACP: 1.7–6.6–10.9 s). The fPL/ACP eliminated nasopharyngeal reflux. Maximum tongue pressure increased from 10.6 to 15.3 kPa. The PAS score was 1 point, with no findings of penetration or aspiration.

The effects of the fPL/ACP on speech function are presented in **Table 2**. The intelligibility for the 100 Japanese phenomes increased from 68.6% to 73.3% and that for the 100 words increased from 95.5% to 98.5%. The MPT increased from 10.7 to 12.8 s.

The patient had temporarily elevated CRP from day 122 because of aspiration pneumonia, which improved with antibiotic treatment without nil per os. Following combined treatment with the fPL/ACP, swallowing and physical training, and nutritional management, the patient was discharged on day 216. At the time of discharge, she was able to have soft and bite-sized foods and her Alb level had improved to 4.0 g/dL. Although the BMI showed no change at 13.0 kg/m^2, physical function improved with a grip strength of 11.4 kg. Tongue pressure measured without fPL/ACP was 10.6 kPa.

This case report was approved by the Seirei Mikatahara General Hospital Ethics Committee (protocol approval no. 21–4, approval date April 16, 2021). The patient provided written informed consent for publication of this report.

DISCUSSION

In the present case, use of the fPL/ACP showed positive effects on dysphagia and dysarthria caused by DLBCL and sarcopenia. To the best of our knowledge, this is the first report to demonstrate the quantitative effects of integrated PAP and PLP, especially for dysphagia.

The benefits obtained from the fPL/ACP were a combination of its effects as a PAP and an f-PLP. As the PLP component, the fPL/ACP prevented nasopharyngeal reflux and provided more force to push the food toward the oropharynx and hypopharynx. It also improved hypernasal speech and speech intelligibility. As the PAP component, the fPL/ACP improved bolus transit, increased tongue pressure, and allowed clear articulation of consonants. The reduction in the pharyngeal residue cannot be attributed to only one component of this device. Many factors were involved in pharyngeal residue, including pharyngeal contractile muscle activity, tongue pressure, UES opening, and pharyngeal swallowing pressure. The combined effects of PAP and PLP, such as improved tongue pressure and prevention of nasopharyngeal reflux, led to a decrease in pharyngeal residue. It has been considered that use of a PLP may inhibit swallowing,[7,9] although evaluation of the f-PLP developed by Ohno et al.,[8] which featured a flexible lift, showed that it reduced discomfort during swallowing. In the present case, the fPL/ACP, which is a combination of PAP and f-PLP, demonstrated the positive effects of both components on swallowing function. We believe that fPL/ACP can be considered among the prosthetic modalities used in the management of dysphagia and dysarthria.

In the present case, the cause of dysphagia and dysarthria remain unclear. DLBCL causes cranial neuropathy and induces neurogenic dysphagia.[16,17] In this case, contrast-enhanced magnetic resonance imaging did not reveal cranial nerve involvement. However, vagus nerve palsy caused by DLBCL has been reported previously, and the soft palate dysmotility in the present case may have been caused by a similar mechanism.[18] Sarcopenia refers to the loss of muscle mass throughout the body associated with aging and malnutrition. It also affects the pharyngeal muscles.[19] The patient's dysphagia was thought to be attributed to the combined effects of the impaired tongue and soft palate motility caused

by DLBCL-derived cranial nerve impairment and weakened pharyngeal contraction caused by sarcopenia.

The improvement of swallowing function in this case was not attributed to the use of the fPL/ACP alone. Comprehensive treatment, which included administration of an fPL/ACP, nutritional support from the perspective of rehabilitation nutrition, swallowing training by an SLHT, and physical and occupational therapy to improve physical function, helped to improve the patient outcome.

In determining the indications for fPL/ACP, it is important to assess motor deficits of the tongue and soft palate through VFSS and dysarthria evaluation. They are caused by various diseases, including neurological disorders (i.e., cerebrovascular and neuromuscular diseases), and tissue loss after head and neck cancer surgery. If the causative disease is a condition for which recovery is expected, such as acute cerebrovascular disease, the prosthesis is fabricated for two purposes: as a compensatory prosthesis and as a training tool. PLP and PAP are expected to not only compensate for dysfunction, but also to encourage improvement in soft palate and tongue function, respectively, with continued use. Patients who achieve good functional improvement may no longer need to use the prosthesis. Concerning this patient, the symptoms may change negatively during the long-term course of DLBCL. Therefore, we basically recommended continuous use of the prosthesis. However, the symptoms of sarcopenia may be improved with an improved nutritional status.

Some patients show discomfort or reject the use of a prosthesis. In our case, the patient commented on the ease of eating, but conversely, there were negative reactions, such as difficulty in tasting and sensory disturbance. To encourage the continued use of a prosthesis, it is critical to convey the significance and benefits of the device to the patient. Therefore, patients with dementia or other cognitive impairments may exhibit lower compliance. In the present case, quantitative measurements, such as the NRRS and speech intelligibility test, demonstrated the effectiveness of the fPL/ACP. These evaluations were helpful in reinforcing the value of this device to the patient. The rehabilitation physician, dentist, and SLHT followed up with the patient to facilitate continued use of the prosthesis after discharge, and the dentist adjusted the prosthesis when necessary.

To date, the treatment of dysphagia and dysarthria with an intraoral prosthesis is relatively uncommon. However, dentures, which are more widely used than PAP and PLP, also improve swallowing function.[20] Prosthetic treatment improves speech and swallowing by altering the structure of the oral cavity. In clinical practice, we often encounter cases of dysphagia caused by complex factors, as in this case. To maximize the effectiveness of the intraoral prosthesis, patient rehabilitation through transdisciplinary collaboration is important.

CONCLUSION

In the present case, fPL/ACP, an integrated PAP+PLP device, improved dysphagia and dysarthria caused by DLBCL and sarcopenia. The fPL/ACP reduced pharyngeal residue, shortened bolus transit time, and improved speech intelligibility. This device has the combined benefits of PAP and PLP and reduces discomfort during swallowing. Therefore, it can be considered a potential treatment modality for patients with tongue and soft palate dysmotility.

CONFLICTS OF INTEREST

The authors declare no conflict of interest.

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