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A novel anatomy-based five-points eight-line-segments technique for precision subtotal tongue reconstruction: A pilot study

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ABSTRACT

Objectives: A no-flap design based on actual tongue anatomy has been reported to achieve precise reconstruction, but large carcinologic tongue defects remain extremely challenging to treat. The aim of this pilot study was to explore an anatomy-based flap design for precision subtotal tongue reconstruction.
Material and methods: Thirty-five patients were randomly divided into two groups, a “five-points eight-line-segments” (FIPELS) technique flap design group (18 patients) and a conventional surgery group (17 patients), to undergo subtotal tongue reconstruction. All patients received an anterolateral thigh flap with nerve coaptation to the lingual or glossopharyngeal nerve. Swallowing function, speech intelligibility, and cosmetic results were assessed with a Likert scale, and epicritic and proprioceptive sensitivity and thermosensation of the neotongue were investigated. Outcomes were analyzed, and a p-value < 0.05 was considered significant.
Results: The FIPELS flaps were significantly larger than conventionally designed flaps. All flaps healed uneventfully, except in two patients who presented with partial necrosis. Relative to patients in the conventional group, patients in the FIPELS group showed significantly improved swallowing function ($p = 0.043$) and cosmetic results ($p = 0.017$) but not speech intelligibility ($p = 0.154$). Patients in the two groups recovered comparably in terms of epicritic and proprioceptive sensitivity and thermosensation.
Conclusion: The anatomy-based FIPELS technique is an innovative and effective solution for subtotal tongue reconstruction.

Introduction

Tongue cancer is the most common oral malignancy, and surgical dissection and reconstruction have become the most accepted treatments for oncological control. We commonly use the group classification proposed by Urken [1] to classify defects after ablation. Guidelines and techniques for tongue reconstruction have been previously presented [2–10]. Generally, for small defects or hemiglossectomy, no reconstruction is necessary to maintain the remaining tongue function; otherwise, reconstruction using a pedicle or free flap should sustain the mobility of the remaining tongue. However, subtotal or total tongue defects, which are produced by transverse resection of this organ, have the worst functional results and are also a challenging issue in reconstructive surgery.

The processes of mastication, deglutition, and phonetic articulation are all actively mediated by the interaction of the neotongue with other

Abbreviations: FIPELS, five-points eight-line-segments; ALTF, anterolateral thigh flap; 3D, three dimensional; TM, total mobile tongue; TMB, subtotal tongue and base (up to 2/3 of the base of the tongue); TMB 1/3, subtotal tongue and 1/3 of base; TMB 2/3, subtotal tongue and 2/3 of base; TPD, two-point discrimination test

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oral cavity structures, such as the palate, floor of the mouth, and dental arches. Thus, when performing reconstruction of defects caused by subtotal or total glossectomy, it is important to allow for adequate folding of the skin paddle to obtain proper volume and protuberance of the neotongue. Regarding functional anatomical restoration, various reconstruction methods have been developed and reported to achieve functional outcomes [2–5,11,12]. However, even with these methods, achieving a sufficient protuberance that can imitate the natural tongue in the oral cavity remains difficult, and currently, the flap size used for certain types of defects is based only on the surgeon's experience. Therefore, a better technique for functional anatomical reconstruction would be to dissect and unfold the tongue based on anatomical landmarks, which can be mapped as a geometric configuration, to assist in flap design.

In this study, we used virtual surgical planning for the dissection of the subtotal tongue and to address flap design, which we based on anatomical landmarks and named the "five-points eight-line-segments" (FIPELS) technique. We compared the functional and cosmetic outcomes of the FIPELS technique with conventional surgery for subtotal glossectomy reconstruction.

Material and methods

Between May 2014 and August 2016, 37 patients with primary squamous cell carcinoma (T3 or T4) according to 7th edition American Joint Committee on Cancer (AJCC) staging system were enrolled and randomly assigned to FIPELS (n = 18) or conventional surgery (n = 19). Two participants (conventional group) withdrew consent immediately after randomization: one because of financial burden and one who elected to pursue Traditional Chinese Medicine. These patients were therefore lost to follow-up. Thus, 35 consecutive patients underwent subtotal glossectomy and completed follow-up. The mean age of the 35 patients was 55.5 years (range, 28.9–77.0 years), and twenty-three male and twelve female patients were included. The tumors were classified as T3 for nineteen patients and as T4 for sixteen patients. Total mobile tongue (TM) resection was performed in fifteen patients, whereas the remaining twenty patients underwent resection of the subtotal tongue and base (TMB), including nine patients with resection of the subtotal tongue and 1/3 of the base (TMB 1/3) and eleven patients with resection of the subtotal tongue and 2/3 of the base (TMB 2/3) according to Urken et al.'s classification [1]. There was no attempt to control for stage (T3/T4) or defect type during randomization. All patients underwent reconstruction with an anterolateral thigh flap (ALTF), and a tracheotomy and gastric tube were used during the operation. This study was conducted in accordance with the principles of the Declaration of Helsinki, and informed consent was obtained from those who participated. The sample size and surgical procedures were approved by the Ethics Committee of Sun Yat-Sen Memorial Hospital.

Anatomy of the hemitongue

The anatomy of the tongue must be meticulously studied in order to achieve an anatomic flap design. To the best of our knowledge, no previous work has examined the conversion of the tongue contour from a three-dimensional structure to a planar graph. We used virtual surgical planning for the dissection of the subtotal tongue. First, we performed magnetic resonance imaging (MRI) to acquire images of an adult volunteer's tongue and to segment the subtotal tongue, according to landmarks, using ProPlan CMF software (Materialise, DePuy Synthes, Belgium) (Fig. 1). The resection margin comprised five landmarks, including the apex of the tongue (A), frenulum of the tongue to the sublingual caruncle (B), mucosa and gingiva of the floor of the mouth to the retromolar pad (C), terminal sulcus to the foramen cecum of the tongue (D), and posterolateral corner of the tongue base (E). We then incised the hemitongue specimen between the superior longitudinal muscle and the deep muscles, including the transverse and vertical muscles, while leaving the lateral border of the specimen intact to act as a hinge (Video 1). Thus, the dorsal (superficial) portion contained the superior longitudinal muscle and its overlying mucosa, whereas the ventral portion contained the transverse muscle, vertical muscle, inferior longitudinal muscle and several extrinsic muscles, including the genioglossus, hyoglossus, and styloglossus. After the hemitongue was incised, the planar graph resembled an irregular pentagon composed of five points (A, B, C, D and E) and six sides (A-B, B-C, C-D, D-E, A-D, and A-E). Side A-E represents the lateral border of the tongue, separating the dorsal hemitongue width (a) from the ventral width (b). Therefore, we called this method the five-points eight-line-segments (FIPELS) technique and used it to tailor the hemitongue and the TM. To avoid

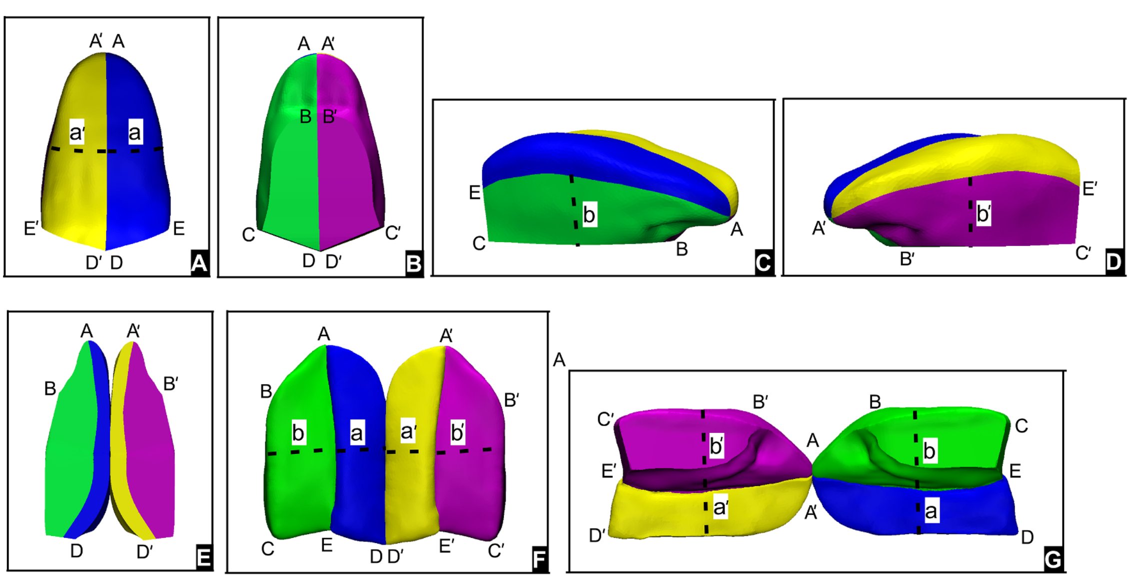


Fig. 1. Segments of the subtotal tongue (TM) using a three-dimensional virtual technique. The left and right mobile tongues were dissected using the FIPELS technique.

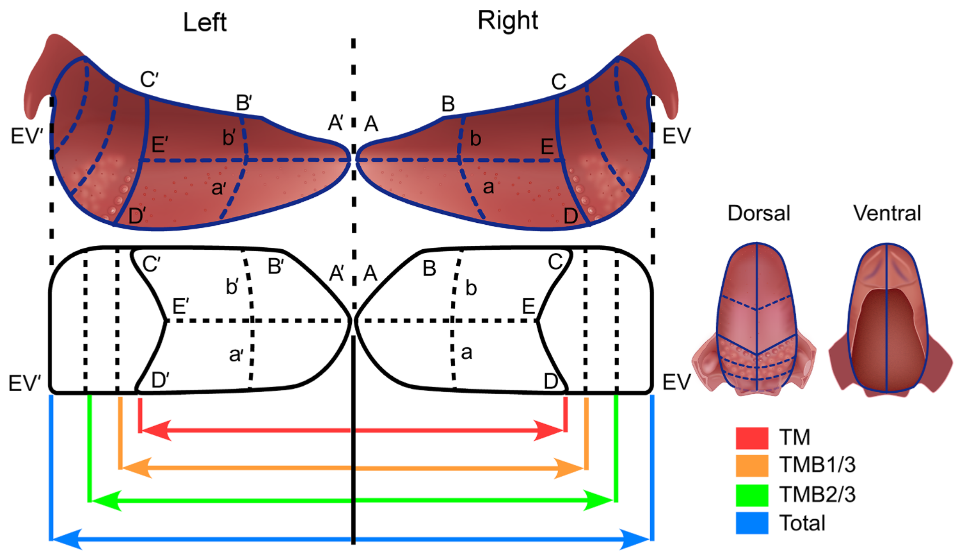


Fig. 2. A schematic illustration of four types of standardized flap designs for subtotal/total tongue reconstruction based on the FIPELS technique.

tension at the donor site, we used the kiss technique [13] to separate both hemitongues along the midline and rotate them 90°. Finally, the subtotal/total tongue was divided into two irregular pentagons (Fig. 1).

To achieve an accurate flap design, flap size was determined based on the FIPELS technique. However, it was impossible to measure the subtotal or total tongue because of the deformation caused by advanced disease (T3 or T4). Thus, we collected the tongue size of the TM and TMB in 20 adult patients (without tongue disease) who underwent parotid gland surgery. Considering that flaps shrink after harvesting, the tongue was positioned near the incisors when the patients were under general anesthesia to compensate for the potential contraction. Then, a flexible ruler was used to measure the size of the tongue, based on the FIPELS technique, as well as the length between (D) and the epiglottic vallecula (EV) (Table S1). Finally, the size of the subtotal/total neotongue was standardized into four types of designs, including TM, TMB 1/3, TMB 2/3 and the total tongue (Fig. 2).

Supplementary data associated with this article can be found, in the online version, at https://doi.org/10.1016/j.oraloncology.2018.12.004.

Operative technique

Flap design and harvesting

All procedures were performed with a two-team approach. One group performed tumor ablation and neck lymph node dissection, while the other team harvested the flap. For ALTF harvesting, flap design began with conventional mapping of perforators from the descending branch of the lateral circumflex femoral artery with a Doppler probe. For the FIPELS group, flaps for TM and TMB defects were classified and determined according to the average size based on the normal tongue (Fig. 3). Suprafascial dissection was performed conventionally by identifying the previously mapped perforators. For patients who did not present with clear perforators during intraoperative macroscopic

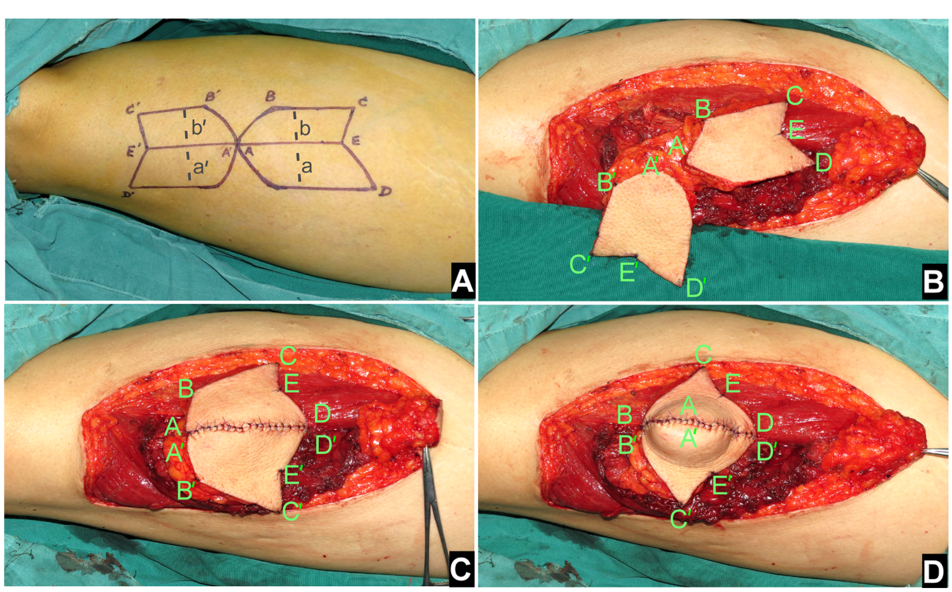


Fig. 3. The flap design and the ALTF harvested using the FIPELS technique for subtotal tongue (TM) reconstruction. The flap was harvested based on the average size of a healthy subtotal tongue. The presutured ALTF used in the FIPELS technique after pedicle skeletonization is shown.

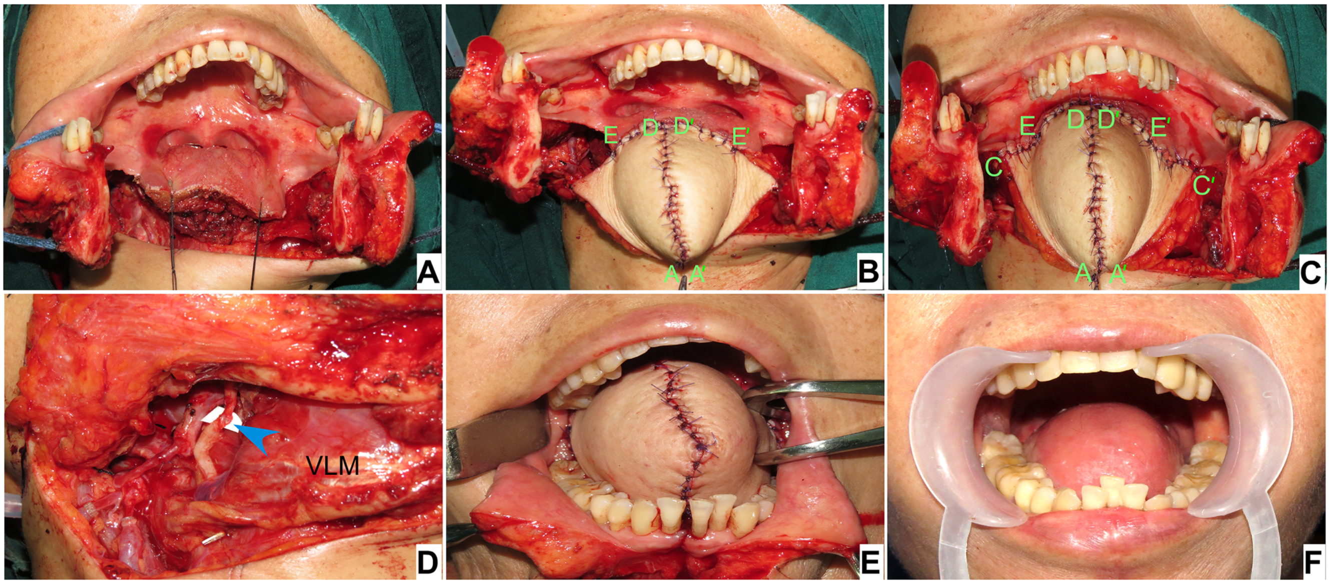


Fig. 4. Reconstruction of a subtotal tongue (TM) defect using an ALTF and the FIPELS technique. An intraoperative view of a subtotal tongue defect after tumor ablation. The ALTF was sutured with the remaining tongue tissue and the mucosa in the floor of the mouth based on the FIPELS landmarks. The arrows indicate nerve coaptation (the cutaneous nerves to the lingual nerves). The mobile neotongue is shown after reconstruction, intraoperatively and at twelve months postoperatively.

evaluation, we harvested a portion of the vastus lateralis along with its entire deep fascia to preserve blood flow. Some subcutaneous layers and the vastus lateralis were included in the skin paddle to fill the dead space. However, subcutaneous adipose tissue with a thickness of more than 1.5 cm was trimmed in overweight or obese patients. Additionally, a sensory branch from the lateral femoral cutaneous nerve was dissected proximally up to the anterosuperior iliac spine and was included in the flap to allow sensory recovery of the neotongue. After complete pedicle skeletonization, the two irregular pentagons were folded and sutured along A-D (A-EV) and then A-B to recreate the whole body and tip of the neotongue. Blood perfusion was also examined before the flaps were transferred to the defect sites (Fig. 3, Video 1).

Table 1
Patient demographics, operative data and follow-up.

	FIPELS group			Conventional group			p
	TM (n = 7)	TMB (n = 11)	p	TM (n = 8)	TMB (n = 9)	p	
Sex, male: female	5:2	7:4	0.572	5:3	6:3	0.627	0.592
Age, mean (range, years)	53.6 (34.3–73.4)	57.2 (41.5–75.8)	0.632	55.8 (38.9–69.2)	54.7 (28.9–77.0)	0.767	0.832
Stage, T3:T4	6:1	4:7	0.057	6:2	3:6	0.109	0.573
pN +	2	4	0.572	2	3	0.563	0.546
Defect type (TMB 1/3: TMB2/3)	–	5:6	–	–	4:5	–	–
Flap size(cm)	10 × 9	10 × 11.5/ 10 × 13[*]	< **0.001**	8 × 7	10 × 8	< **0.001**	< **0.001**
Nerve coaptation			**0.025**			0.053	0.396
Lingual	7	5		8	5		
Glossopharyngeal	–	6		–	4		
Overall operative time [min (range)]	396.8 (356.3–470.9)	408.7 (350.4–489.5)	0.472	378.9 (342.0–463.7)	395.3 (337.8–498.9)	0.746	0.871
Flap status	All survived	Partial necrosis, 1	–	Partial necrosis, 1	All survived	–	–
Decannulation interval [days (range)]	8 (7–13)	9 (7–12)	0.864	9 (7–13)	9 (7–15)	0.891	0.573
Oral alimentation interval (days)	10 (8–13)	11 (8–13)	0.771	11 (7–13)	11 (9–17)	0.543	0.497
Flap Sensitivity							
Monofilament test (10 g)	All positive	9 positive [#]	–	7 positive	All positive	–	–
TPD(mm)	13.4	16.3[#]	0.291	15.3	15.6	0.674	0.451
Heat-cold test	All positive	All positive [#]	–	All positive	All positive	–	–
ART	2	4	0.572	3	3	0.627	0.592
Median follow-up [months (range)]	29(12–38)	26(10–39)	0.563	28(13–39)	27(12–38)	0.465	0.276
Disease status	All remission	Dead, 2		Dead, 1	Dead, 2		

FIPELS, five-points eight-line-segments; TM, total mobile tongue; TMB, subtotal tongue and base(up to 2/3 of the base of the tongue); TMB 1/3, subtotal tongue and 1/3 of base; TMB 2/3, subtotal tongue and 2/3 of base; TPD, two-point discrimination test ; ART, adjuvant radiotherapy.

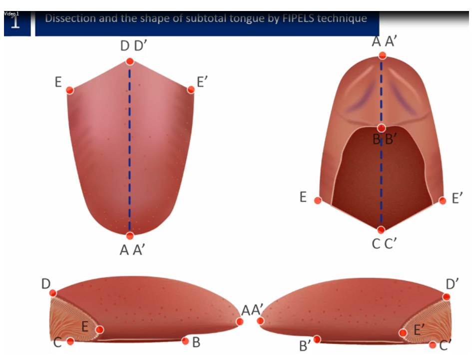
[#] One patient presented with local recurrence within 12 months after surgery, and the flap sensitivity was tested at 10 months postoperatively.

* TMB 1/3 and TMB 2/3 with flap sizes of 10 × 11.5 cm^2 and 10 × 13 cm^2, respectively.

Table 2
Functional and aesthetic outcomes evaluation.

	FIPELS group				Conventional group				P*
	TM	TMB	Total	P#	TM	TMB	Total	P#	
Swallowing				0.417				0.827	**0.043**
Dysphagia	0	0			0	0			
Moderate impairment	0	0			2	2			
Mild impairment	2	5			3	5			
Nearly natural	5	6			3	2			
Speech intelligibility				0.648				0.793	0.154
Tracheostomy required	0	0			0	0			
Unintelligible speech	0	0			1	1			
Acceptable speech	3	5			4	6			
Near normal speech	4	6			3	2			
Cosmetic results				1.000				1.000	**0.017**
Poor	0	0			2	3			
Fair	0	1			2	2			
Good	4	5			3	2			
Excellent	3	5			1	2			

FIPELS, five-points eight-line-segments; TM, total mobile tongue; TMB, subtotal tongue and base(up to 2/3 of the base of the tongue).
Comparisons were performed between the TM and TMB subgroups in the FIPELS or conventional group.
* To evaluate differences between the FIPELS and conventional group, the data from patients in the TM and TMB groups were combined.



Video 1. Imitation of the dissection and the shape of the subtotal tongue using the FIPELS technique.

In the conventional group, the flaps were designed as ellipses or spindle-shaped pieces, with stable sizes of 8 × 7 cm^2 and 10 × 8 cm^2 for TM and TMB defects, respectively. Then, flap harvesting and sensory branch dissection were performed as in the FIPELS group. The donor site was directly closed without skin grafts in both groups.

Subtotal tongue reconstruction

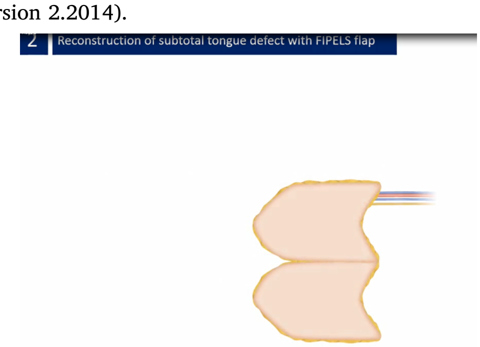
After the flap was harvested and transferred to the oral cavity defect, end-to-end anastomosis of vessels and nerve coaptation (the cutaneous nerves to the lingual nerves or glossopharyngeal nerve) were performed in both groups. Then, the presutured FIPELS flaps were located on the residual tongue and lining of the floor of the mouth according to the FIPELS landmarks. Finally, a protuberant neotongue was produced (Fig. 4, Video 2). For the conventional group, the flap inset was completed to cover the defect without the FIPELS landmarks.

Outcome evaluation

A Likert scale was used to assess swallowing safety and efficiency, speech intelligibility, and cosmetic results at 6 months after surgery (Table S2) [5]. Swallowing safety and efficiency were assessed by two independent physicians based on the videofluoroscopic swallow study (VFSS). Speech intelligibility and cosmetic results were evaluated by two trained speech pathologists and two independent plastic surgeons who did not join the study and were blinded to the surgical procedure. Tests of epicritic and proprioceptive sensitivity of the neotongue were conducted 12 months postoperatively by two independent neurological physicians who were blinded regarding nerve coaptation. We used a basic hand set of Semmes-Weinstein monofilaments with 10 g to assess proprioceptive sensitivity, and a two-point static discriminatory test was performed using a compass with a blunt or sharp-pointed tip; thermosensation was assessed using a tube filled with hot (50 °C) or cold (4 °C) water [5]. The Fisher's exact test and Kruskal-Wallis test were used for comparisons between the two groups. Statistical analyses were performed using the SPSS 18.0 package (SPSS, Chicago, IL, USA), and a p-value of < 0.05 was considered statistically significant.

Results

The age, sex, and other clinical characteristics of the two groups were comparable (Table 1). No flap failure occurred, except for two cases of partial necrosis in which minor wound dehiscence at the floor of the mouth or the base of tongue occurred; suture revision was required but did not compromise the final functional outcome. The mean time to decannulation and the oral alimentation interval were comparable between the FIPELS and conventional groups as well as within groups. However, the flap size was significantly larger in the FIPELS group than in the conventional group, and the flap size was significantly larger for TMB defects than for TM defects in both groups ($p < 0.001$). All the resection margins were negative, and patients with pN+ (n = 11) and vascular embolism (1 in the conventional group) received adjuvant radiotherapy according to the NCCN guidelines (version 2.2014).



Video 2. Reconstruction of the subtotal tongue (TM) using an ALTF with the FIPELS technique.

Video 3. At the 6-month follow-up, a VFSS demonstrated effective bolus propulsion without aspiration in a patient who underwent the FIPELS technique using an ALTF for subtotal tongue (TM) reconstruction.

All patients recovered sensitivity of the neotongue, except for two patients with TMB defects in the FIPELS group and one with a TM defect in the conventional group (negative sensitivity for 10 g using Semmes-Weinstein monofilament); however, thermosensation was present in all patients, and no significant between-group or within-group differences were observed for the two-point discrimination test (TPD). In the functional and cosmetic outcome evaluations (Table 2), significant differences were observed between the FIPELS and conventional groups regarding swallowing safety and efficiency (Video 3 demonstrates effective bolus propulsion without aspiration in a VFSS) ($p = 0.043$) and cosmetic evaluation ($p = 0.017$) but not speech intelligibility ($p = 0.154$). No significant difference was found regarding TM and TMB defects in each group.

Discussion

Improving long-term survival and quality of life of patients have recently received increased attention, and functional recovery has been increasingly emphasized. The high success rate of free flaps has revolutionized the ability to achieve more pliable and reliable flaps that are ideal for ensuring a certain degree of movement with a reduction in donor-site morbidity [2,5,13]. However, tongue reconstruction, particularly for subtotal or total tongue defects, remains a highly specialized area for reconstructive surgeons. Because the tongue is important for several activities, such as speech, deglutition and airway protection, surgeons have sought to develop multiple strategies to restore the neotongue due to its great mobility and interaction with other oral cavity structures, such as the palate, floor of the mouth, and dental arches [4,5,14,15]. Most of these designs are intended for hemiglossectomy defects; in fact, little information has been reported regarding the refinement of total or subtotal tongue reconstruction [16–19]. For total tongue defects, Engel et al. [20] designed a pentagonal anterolateral thigh myocutaneous flap that facilitates better flap inset, adequate volume, and an aesthetically pleasing neotongue tip but does not allow adequate forward and lateral propulsion of the tongue. Leymarie et al. [2] described a modified flap design, called the “cathedral triptych”, to recreate a near normal neotongue shape, with a more projected tip and vertical bulk. However, the tip and the body of the neotongue are bulky and limit tongue mobility. Recently, Longo et al. [5] developed a mushroom-shaped ALTF that allows for simultaneous tongue and floor-of-mouth reconstruction and restores a projected tip and vertical bulk that can contact the palate. However, such a conformation does not seem to produce a natural shape that can allow a certain degree of interaction with the dental arch and achieve lateral propulsion.

To pursue a precise three-dimensional configuration of flap design and molding and to restore the neotongue as an effective organ, we suggest that the base must have an exact dimensional match between the flap and tongue defect. In a review of the literature [2,3,5,20], we found that previous flap designs focused on only the general contour of the tongue, which could not be quantified and was underestimated. Thus, these designs did not achieve desirable matches. No study has reported using the anatomy of the tongue as a guide to shape the flap. In the present study, we performed dissection of the subtotal tongue to achieve the translation of a two-dimensional image to a three-dimensional representation though virtual surgical planning. The dissection was based on the anatomical landmarks of the tongue and was named the FIPELS technique. In the FIPELS technique, the shapes of TM and TMB are different from previous designs [2,3,5,20]. The neotongue comprises two irregular pentagons, while the width (a and b), length (A-D/A-EV, A-E and B-C), frenulum of the tongue (A-B) and posterior attachment (D-E and E-C) of the neotongue are exact replicas. Based on the configuration of a healthy tongue, we found that the standardized flap size of TM or TMB defects in the FIPELS group was larger than that in the conventional group or in other studies [2,5]. The FIPELS technique restores a near-natural mobile tongue shape and volume, which increases the contact of the neotongue with the palate, cheek and teeth in the oral cavity. This technique directly affects functional and aesthetic results. We used the FIPELS technique to separate the subtotal/total tongue into two lobes, which can reduce dehiscence at the donor sites; all donor sites underwent primary closure without any complications, even for TMB2/3 defects for which the total flap size was as large as $10 \times 13\,cm^2$.

Functional outcomes for swallowing and cosmetic appearance in the FIPELS group were significantly better than those in the conventional group, which confirmed that the FIPELS design can be safely and effectively applied for subtotal tongue reconstruction. Although a fully functional tongue is unlikely to be achievable for TM or TMB defects, the FIPELS technique can reproduce a protuberant and bulky yet mobile neotongue with improved deglutition, better processing of the food bolus and oral clearance. A videofluoroscopic swallowing study showed good neotongue motility, with good contact to the anterior palate and adequate posterior elevation and sealing of the pharyngeal sphincter without evidence of aspiration. However, we did not identify a significant difference between the FIPELS and conventional groups when evaluating speech intelligibility in the current study. Our results are consistent with those of a previous study by Jeong et al. [4], who reported that the preservation of flap volume is important to achieve superior swallowing capacity but insufficient to promote speech outcomes in total tongue reconstruction. However, Kimata et al. [21] reported that sufficient flap bulk promotes speech intelligibility. Certain issues may contribute to these contrasting results. First, the tongue interacts precisely with nearby structures, such as the teeth, alveoli, and palate, during pronunciation [22]. Second, it is technically impossible to recreate a multidirectional mobile neotongue; however, the establishment of only one vector of tongue movement may impair meticulous tongue motion characteristics during normal speech.

There is controversy regarding the use of reinnervated flaps [23–28]. Researchers have found that innervated flaps lead to a higher satisfaction rate [25], can maintain the symmetry of the reconstructed tongue [24] and promote faster recovery of two-point discrimination, thereby potentially avoiding tooth bites and burn injury to the flaps [5,24]. However, no significant effect of reinnervated flaps on swallowing function was observed during long-term follow-up evaluations [24,25]. In the present study, we performed nerve coaptation on all patients, and flap sensitivity was recovered in all patients in both groups, except three patients who were negative for the monofilament test. However, no significant differences were observed between the FIPELS group and the conventional group in the other evaluations of flap sensitivity. We believe that the improved swallowing and cosmetic outcomes of the FIPELS group may have been due to the flap design rather than the reinnervated flaps. However, the effects of reinnervated flaps need to be demonstrated by prospective studies and studies with an extended follow-up period.

There are limitations to the current study, including the small cohort size, the discrepancy in the group cohorts and the short follow-up duration. The present cohort included patients with late stage disease, and the tongue deformation prevented a normal tongue size from being achieved. Therefore, we designed the flap sizes in the FIPELS group based on an average configuration of a healthy tongue. We could not exclude discrepancies between average flap size and individual oral cavity volumes or dental arches, which may have impaired the functional and cosmetic outcomes. The average tongue size may vary among populations. The association between tongue size and oral cavity volume must be identified in future studies, and individual flap size may be calculated based on radiography. In addition, we used a very standardized flap design for subtotal tongue defects; further adjustment may be considered in future studies since some defects do not involve all intrinsic and extrinsic muscles on both sides and tongue resection is not symmetric. Notably, the FIPELS technique could be popularized for hemitongue reconstruction, as flap size can be accurately measured based on the remaining healthy hemitongue. Although the FIPELS technique can significantly improve functional and cosmetic outcomes of subtotal tongue reconstruction, techniques for functional muscle reconstruction and reinnervated muscle to create a multidirectional mobile neotongue must be further explored.

In summary, the anatomy-based FIPELS technique is an innovative and effective option for subtotal tongue reconstruction. Its precise design enables the reproduction of a neotongue with a normal shape and volume, and it yields predictable and reproducible results. Because it defines the defect based on anatomical landmarks and line segments, guaranteeing an exact match with defects, we believe that the FIPELS concept is not only applicable for subtotal or total tongue reconstruction but can also be used for other applications.

Acknowledgments

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Conflict of interest statement

The authors declare no conflicts of interest.

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