Primary Pharyngeal Flap With Palate Repair Improves Speech Outcome in Older Children and Adults

A Comparative Study

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Background: Repair of cleft palate after 6 years of age is controversial in regard to the surgical procedure and the speech outcome. Primary repair alone may not be considered sufficient to achieve intelligible speech. The authors consider addition of pharyngeal flap at the time of primary repair to be a significant factor in improving speech.
Methods: Prospectively maintained data of all cleft palate patients operated from 2013 to 2017 (5 years) was analyzed to identify patients older than 6 years. Complete cleft palate, incomplete cleft palate, and cleft of the soft palate were further stratified according to different Randall types. They were divided into 2 main types: primary palate repair only and primary palate repair with pharyngeal flap. Speech was assessed preoperatively and 12 months postoperatively by Pittsburgh weighted speech scale.
Results: A total of 139 patients were analyzed. There were 78 males and 61 females. Their ages ranged from 6 to 60 years (mean age, 12.5 years). The overall preoperative speech score in palate repair–only group was 12.15, whereas the postoperative score was 7.32. In patients who underwent primary pharyngeal flap along with palate repair, the preoperative speech score was 11.3, and the postoperative score was 3.76.
Conclusions: In select group of patients who report late for palate repair, addition of pharyngeal flap along with the primary palate repair improves the speech outcome in all Randall groups.

Key Words: cleft palate repair, cleft palate with pharyngeal flap, delayed cleft palate repair, late palate repair, primary pharyngeal flap, velopharyngeal insufficiency

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Intelligible speech is one of the main determinants of success of cleft palate surgery.[1] Velopharyngeal insufficiency is one of the major causes of unintelligible speech after cleft palate repair.[2] A number of techniques have been devised to address the problem. Levator dissection and retropositioning[3] to pharyngeal flaps[4] and sphincter pharyngoplasty[5] all improve the velopharyngeal function. In developed countries, the health care systems are robust, and repair of cleft palate is completed within 2 years of birth so that proper speech development should occur.[6] However, there are many countries in the world where patients with cleft lip and palate are not operated on until early childhood because of the lack of proper health care facilities.[7] At this stage, a large number of patients have developed compensatory articulation problems,[8] and it is difficult to decide whether palate repair alone or addition of pharyngeal flap or some other procedure will be appropriate for better speech outcome.[9] Many authorities in the world recommend an obturator for this problem.[10] Nevertheless, palate repair at any stage is helpful in improving the speech[11] in addition to providing a barrier between oral and nasal cavities, which is useful for oral hygiene.

Many authorities suggest execution of pharyngeal flap along with primary palate repair if the age of the patient is older than 6 years, especially if the existing palate is short (Randall 3 or 4),[12]; however, scientific evidence on the use of pharyngeal flap with primary palate repair is lacking.[13] The senior author has operated upon a large number of young and adult cleft palate patients and has found that performing a pharyngeal flap at the time of primary palate repair reduces the incidence of velopharyngeal insufficiency, and the patient's speech improves considerably. In addition, the need for second surgery can be avoided. As there is paucity of studies comparing palate repair with or without pharyngeal flap, we decided to carry out a retrospective cohort study that can help us to rationalize the better way of treating this neglected population in which older children and adults have missed the golden time period of speech learning.

MATERIALS AND METHODS

This study was conducted at the Department of Plastic Surgery, Services Institute of Medical Sciences and CLAPP (Cleft Lip and Palate Association of Pakistan) hospital, Lahore. A formal approval was

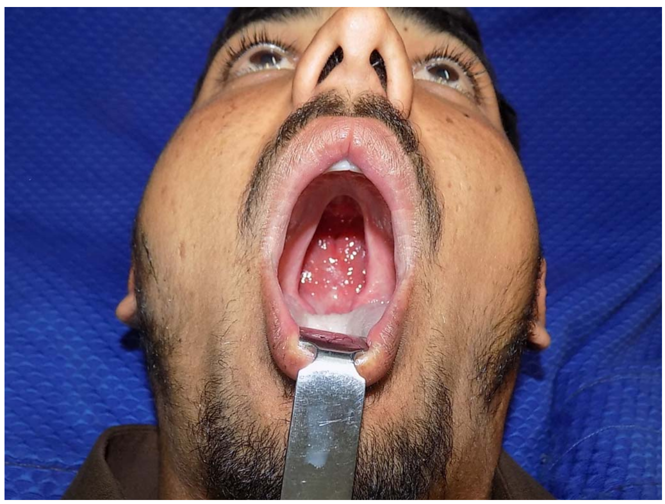

FIGURE 1. Patient with incomplete cleft palate. full color online

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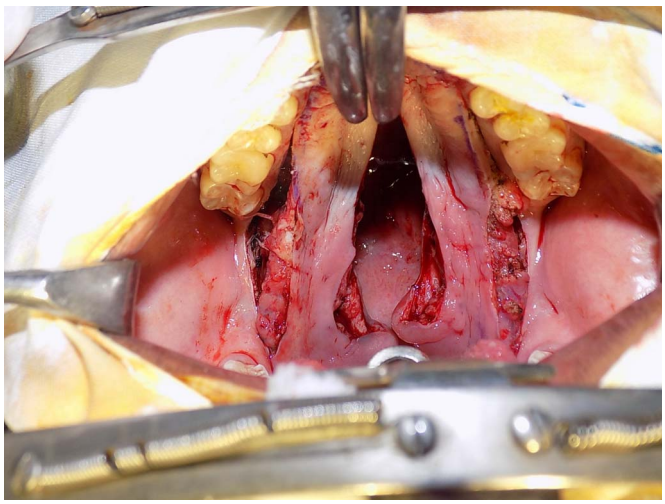

FIGURE 2. Langenbeck procedure in progress. Medial and lateral incisions used to separate the nasal and oral layers along the length of the palate. full color online

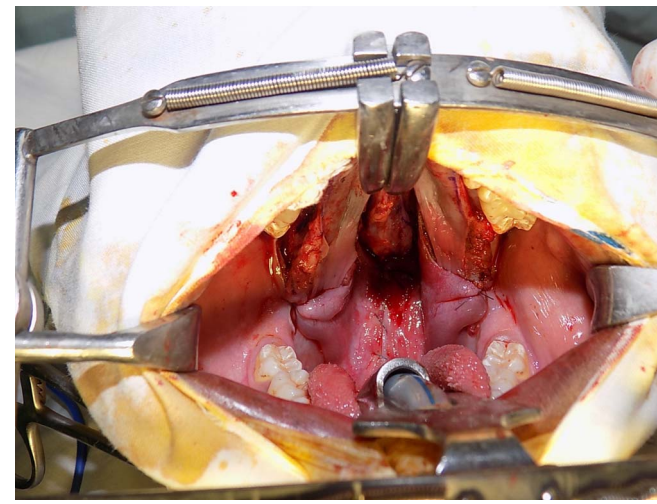

FIGURE 4. Pharyngeal flap raised and turned up to be attached to the nasal layer of the soft palate. full color online

obtained from the ethics committee of the hospital. The prospectively maintained data of 1500 patients (all age groups) over a period of 5 years (January 2013 to December 2017) was searched for primary cleft palate patients 6 years or older (inclusion criteria). Any patient with other congenital anomaly, patients who developed fistula or dehiscence, and patients who could not follow up for 12 months postoperatively were excluded from the study. In the initial study period, palate repair was done without addition of pharyngeal flap, whereas in the later part of the study, the senior author routinely added pharyngeal flap with palate repair. As there are different procedures for complete cleft palate, incomplete cleft palate, and cleft of the soft palate, the patients with complete cleft palate underwent 2 flap palatoplasty (Bardach repair),[14] whereas in patients with incomplete cleft palate and cleft of the soft palate, von Langenbeck palatoplasty[15] was carried out. Levator dissection and retropositioning[11] were carried out in all patients as an essential part of palate repair. In patients of primary pharyngeal flap group, palate repair, levator dissection, and retropositioning, along with superiorly based pharyngeal flap, were carried out. All surgeries were performed by the senior author (G.Q.F.). The way in which the pharyngeal flap was executed is as follows:

After application of Dingman mouth gag and carrying out the dissection of oral and nasal mucosa (Figs. 1 and 2), pharynx was observed for any visible pulsation. Atlas was palpated by the index finger and marked as its base. Pharyngeal flap was marked as V-shaped flap on the posterior pharyngeal wall, based superiorly just caudal to the distal portion of the adenoid pad as shown in Figure 3. Lateral borders of the flap were marked according to the width of pharyngeal wall, and we utilized 60% of the wall, leaving lateral ports equal to 20% on each side. Inferior border was just tapered to "V." The posterior pharyngeal wall was infiltrated with Xylocaine (0.5%) and epinephrine (1:100,000) solution. The width of the pharyngeal flap was never more than 60% of the width of the posterior pharyngeal flap. In the authors' view, this helps to avoid obstructive sleep apnea. The length of the flap should be sufficient enough so that the tip of the pharyngeal flap should reach the anterior-most extent of the split nasal layer of the soft palate. Pharyngeal flap was incised deep to the pharyngeal constrictor muscles

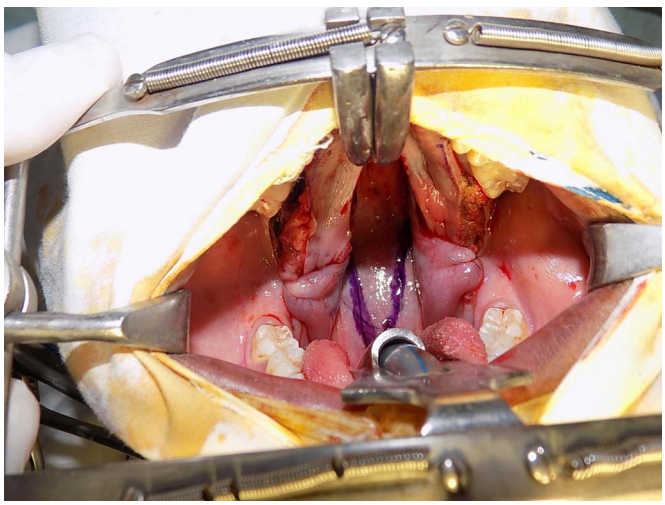

FIGURE 3. Pharyngeal flap marking on the posterior pharyngeal wall. full color online

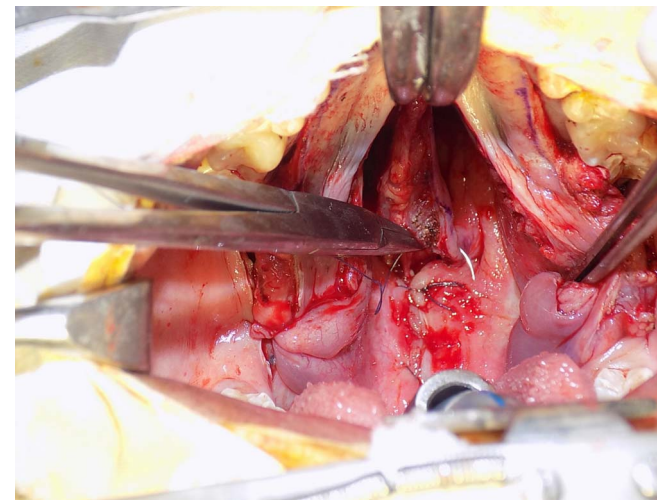

FIGURE 5. Approximation of the left side of the pharyngeal flap with nasal layer of the soft palate on the left side; suture passing through the posterior portion of the pharyngeal flap. full color online

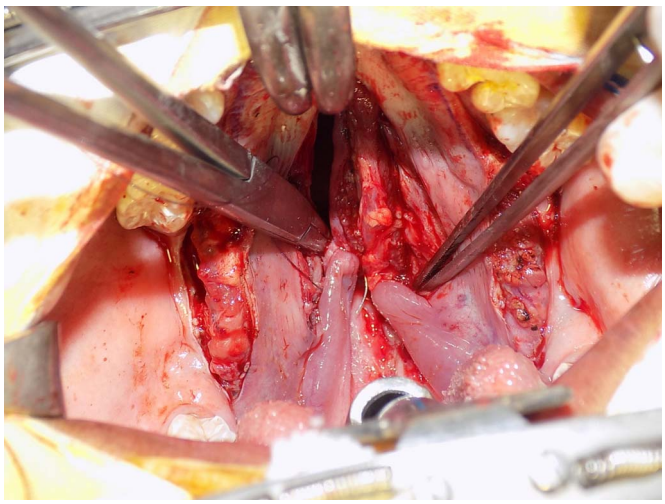

FIGURE 6. Suture being passed from the nasal layer of the soft palate, and then it passes through the posterior portion of the pharyngeal flap on the right side. full color online

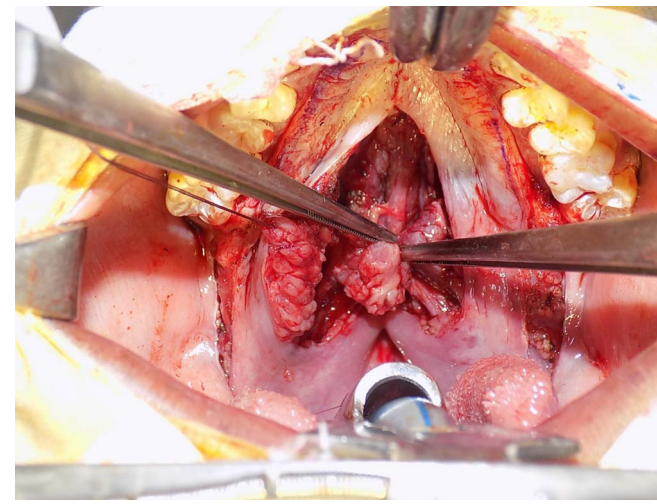

FIGURE 8. Both levators mobilized and transposed posteriorly. First suture will be applied to fix both levators at the base of the pharyngeal flap. full color online

until the glistening fascia was seen. Then, using blunt scissors, the flap was raised off the posterior pharyngeal wall superiorly up to the distal portion of the adenoid pad. Alternatively, one may use monopolar needle cautery to raise the pharyngeal flap.

After the flap is raised, it was turned up for insetting. The tip of the flap was sutured to the anterior-most portion of the nasal layer of the soft palate (Fig. 4). Donor defect at the posterior pharyngeal wall was closed primarily with a running continuous polyglactin 3-0 or 4-0 suture up to the base of the pharyngeal flap. Then, the left nasal layer of the soft palate was attached to the left side of the pharyngeal flap (Fig. 5), and the right side of the pharyngeal flap was sutured to the right nasal layer of the soft palate (Fig. 6). Levator muscles from both sides were identified, transposed posteromedially, and sutured together at the base of the pharyngeal flap (Figs. 7, 8 and 9), taking a good bite of the superior constrictor muscle at the base of pharyngeal flap. Two more sutures were applied (to the levators): one anterior and one posterior to the first key suture. After securing the palatal musculature with the pharyngeal flap musculature, closure of the oral layer was carried out. Uvulae from both palatal flaps were sutured together with vertical mattress suture and then at the base of pharyngeal flap (Fig. 10). A continuous suture of polyglactin 3-0 was used to close the oral layer of the soft palate[16] as shown in Figure 11. Rest of the palate repair was completed in a routine fashion (Fig. 12). Figures 13 and 14 show the 3-week and 6-month postoperative results, respectively.

In summary, there were 2 main groups: group 1, palate repair only (without pharyngeal flap), and group 2, palate repair with pharyngeal flap. In both these groups, the patients with different lengths of the palate were further divided based on the Randall class. In this way, there were a total of 8 groups, 4 for palate repair without pharyngeal flap and 4 for palate repair with pharyngeal flap. The patient is observed for 1 month for complete healing of the palate. Later, the patient is referred to speech language pathologist for speech assessment and therapy. After speech assessment and calculation of speech score, a specific plan is designed for each patient. Patients visited speech pathologists every 2 to 4 weeks for a varying period of 6 to 12 months. Speech was assessed preoperatively and 12 months postoperatively by a dedicated speech therapist who was blinded about the procedure and diagnosis of the patients. Pittsburgh weighted speech score[17] was utilized to assess the speech for nasal air emission, nasality, phonation, and articulation. Data

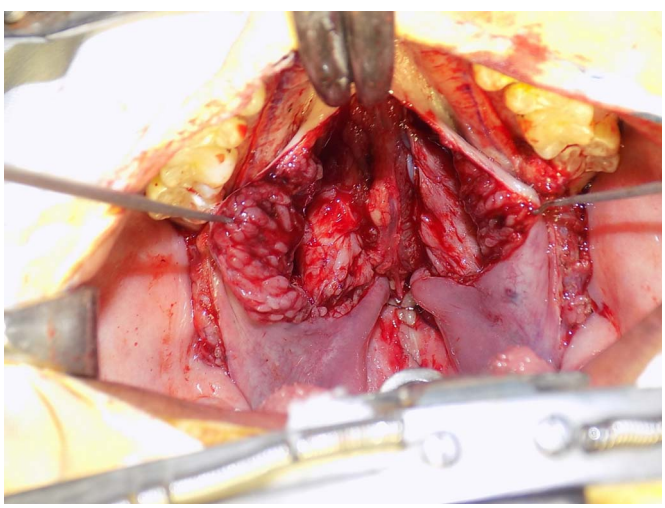

FIGURE 7. Levator muscles dissected on both sides. full color online

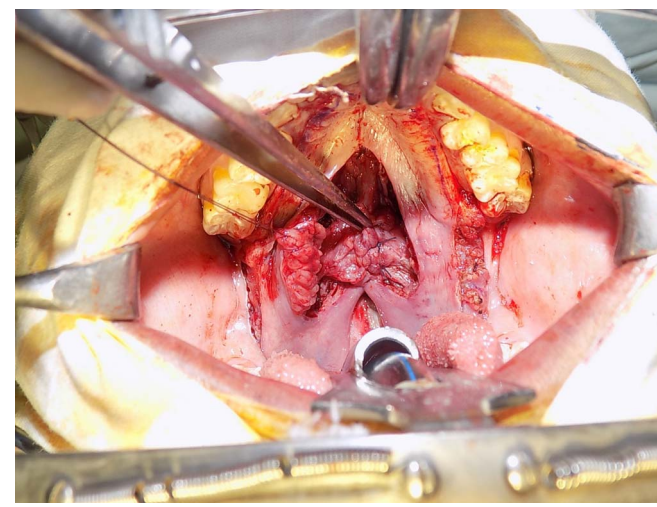

FIGURE 9. Two more mattress sutures applied to both levators, one anterior and one posterior to the first suture. full color online

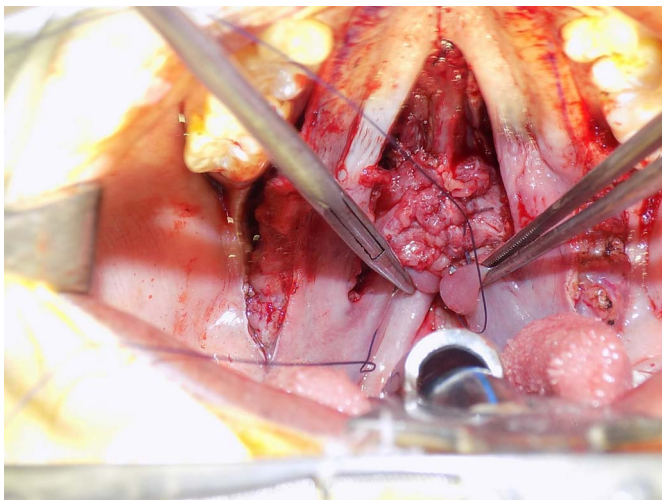

FIGURE 10. Uvulae on both sides fixed to the base of the pharyngeal flap. full color online

entry and analysis were done by using SPSS version 23. Quantitative variables were presented in the form of mean ± SD, and qualitative variables were presented by using frequency and percentages. Independent-samples *t* test was used to compare the speech score in treatment groups; however, paired-sample *t* test was used to see the difference in pretreatment and posttreatment speech scores. Further stratification was done with respect to Randall type to see its effect on speech score before and after treatment. $P < 0.05$ was taken as significant.

RESULTS

A total of 185 patients with age 6 years or older with primary cleft palate were identified. Two patients developed dehiscence, 3 patients developed fistulas, and 41 patients were not able to complete 12 months of follow-up. So, after application of exclusion criteria, 139 patients qualified for data analysis. Their ages ranged from 6 to 60 years, mean age 12.5 ± 7.6. There were 78 males (56.1%) and 61 females (43.9%). The distribution of age, sex, type of cleft palate, operative procedure, preoperative speech score, and postoperative score according to the different Randall class is summarized in Table 1. As none of the patients of

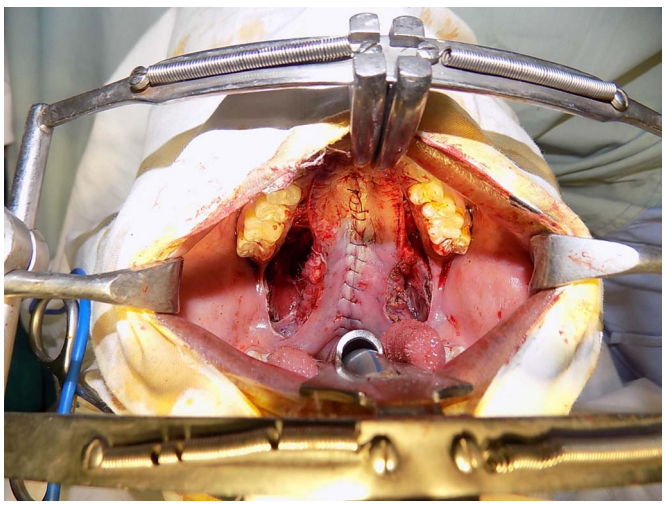

FIGURE 11. Closure of the oral layer and then proceeds anteriorly by a running continuous suture. full color online

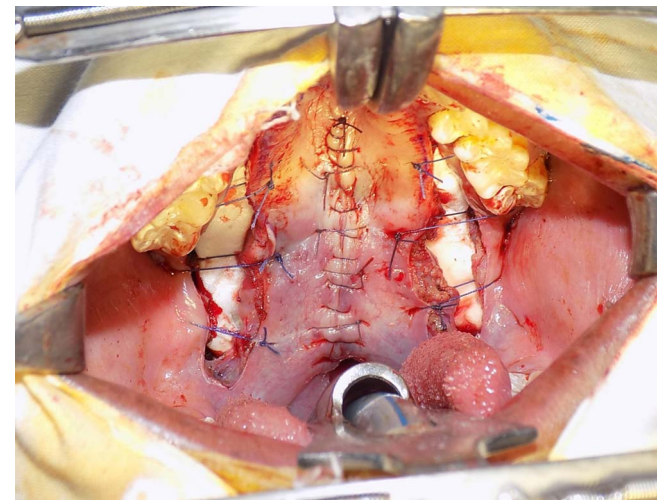

FIGURE 12. Interrupted sutures used to reinforce the midline closure. Lateral defects covered with gelatin foam, secured with interrupted polyglactin sutures. full color online

Randall class 4 qualified for inclusion and exclusion criteria, this class was excluded from the study. The overall preoperative speech score in palate repair–only group was 12.15, whereas the postoperative score was 7.32. In patients who underwent primary pharyngeal flap along with palate repair, the preoperative speech score was 11.3, and the postoperative score was 3.76. The values for different Randall classes' preoperative and postoperative scores along with percentage differences are shown in Table 2. Randall class 3 showed maximum improvement in speech score in patients with primary pharyngeal flap, whereas there was no significant difference in percentage of improvement between different Randall classes in patients in which palate repair was carried out without addition of pharyngeal flap (Table 2). A detailed breakup of different speech parameters among different groups of patients according to Randall's class is shown (Table 3), which shows marked improvement in nasality, nasal air emission, and phonation while not much improvement in articulation.

Regarding the complications, 5 patients developed postoperative bleeding (3 in the palate repair group, whereas 2 in the pharyngeal flap group) for which they were taken to the operating room. Bleeding in 3 patients settled with compression, whereas diathermy was used to

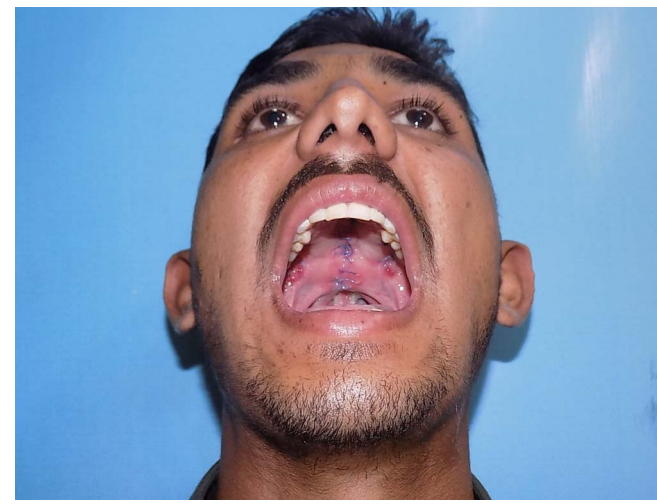

FIGURE 13. Three weeks postoperative. full color online

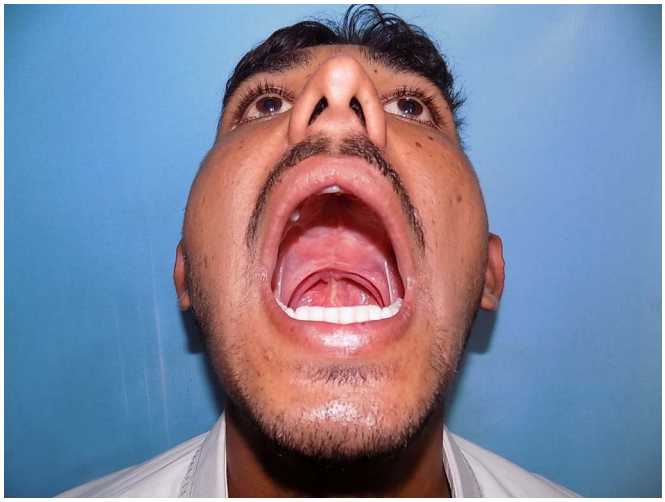

FIGURE 14. Six months postoperative. full color online

control bleeding in the other 2 patients. None of the pharyngeal flap patients needed revision after surgery. Our protocol for assessment of adequacy of nasal airway is to make sure that every patient with pharyngeal flap should be able to breathe spontaneously through the nose with mouth closed. If the patient is unable to breathe normally through the nose, the patient is taken back to operating room to reassess the nasal and airway patency or any faulty placement of sutures causing blockade of lateral ports.

The patients from both main groups who were declared incompetent (score ≥7) after 12 months of follow-up were reassessed by speech language pathologist for velopharyngeal insufficiency and then scheduled for further speech therapy or secondary surgery for velopharyngeal insufficiency.

DISCUSSION

The current study aims at establishing the effects of primary pharyngeal flap surgery in different cleft palate populations on speech. Although speech improvement occurred in all patients regardless of the fact that primary pharyngeal flap or simple palate repair has been carried out. We found that speech improved considerably well in patients in whom pharyngeal flap was added (percentage of improvement 66.72% vs 39.75%). The percentage of improvement seen among different groups also verifies the fact that speech improved more in advancing Randall's class patients. The maximum improvement is evident in Randall 3 patients (71.43%). Nevertheless, even in Randall class 1 patients, the improvement is comparably better in pharyngeal flap patients.

Repair of cleft palate has progressed considerably over the last few decades.[1] Timing of palate repair has profound effect on speech production.[18] In developed countries of the world, the cleft treatment programs are very efficient, and there would be hardly any patient with unrepaired cleft palate beyond 18 to 24 months of age. In developing and underdeveloped countries, the situation is different. A large number of patients with unrepaired cleft palate present to our clinic. At this stage, the patients have developed compensatory articulation mechanisms, which make speech therapy quite difficult. Most of these patients require 6 to 9 months of speech therapy sessions after palate repair, and quite often at the end of these sessions, the speech therapist would suggest pharyngeal flap surgery to increase palatal length and reduce nasal air emission. After execution of pharyngeal flap, the speech improves dramatically. The senior author started performing pharyngeal flaps on patients with short palate 5 years ago and observed improved speech results. Also, the number of speech therapy sessions required to produce intelligible speech was reduced as well.

Recently, Wermker et al.[19] compared speech results of patients undergoing pharyngeal flap, palatal pushback, and a combination of these procedures and reported reduction in hypernasality and improved nasalance scores or subjective speech evaluation. Similarly, Rochlin and colleagues[20] reported an 18-year experience of pushback pharyngeal flap for velopharyngeal insufficiency in 40 patients and found that 91% of the patients achieved adequate velopharyngeal function. Our study showed slightly less improvement of speech scores, but the former studies have not compared different procedures among different Randall types with primary palate repair and in the age range of 6 to 60 years. Also, it may be worthy of note that speech is a complex product of phonation and articulation. If detailed analysis of components of speech is considered, our patients showed remarkable improvement of phonation and hypernasality scores especially among patients in which primary pharyngeal flap was carried out (Table 3). The higher overall scores may be attributable to articulatory errors, which depend on multiple factors in addition to velopharyngeal incompetence.

There is no significant difference in the age of the patients in different groups. Although patients with advancing age tend to develop substitution errors that are more difficult to correct,[8] our study shows that palate repair with or without pharyngeal flap shows definitive improvement in speech in different age groups. It is because of this fact that we advocate palate repair of any patient regardless of age and speech mislearning.

The current literature is scarce on primary pharyngeal flap surgery. Ortiz-Monasterio et al,[21] in 1966, reported that repair of the palate is of minimal benefit after the age of 12 years. Stark and Dehaan[13]

TABLE 1. Demographics of the Patients with Operative Procedures and Speech Scores Among Different Groups of Randall Subtypes

		Randall 1	Randall 2	Randall 3	Total/Mean
No. patients		25 (18%)	98 (70.5%)	16 (11.5%)	139
Age of patients, y		15.2 ± 12.8	12.1 ± 5.9	11.0 ± 4.8	12.5 ± 7.6
Sex of patients	Male	15	56	7	78 (56.1%)
	Female	10	42	9	61 (43.9%)
Type of cleft	Incomplete	13	16	7	36
	Complete	12	59	7	78
	Cleft of soft palate	0	23	2	25
Operative procedure	Palate repair only	13	55	8	76
	Palate repair with pharyngeal flap	12	43	8	63
Speech score	Preoperative score	11.3 ± 1.9	11.8 ± 1.4	12.4 ± 0.7	11.8 ± 1.5
	Postoperative score	6.4 ± 2.9	5.5 ± 3.1	5.7 ± 2.6	5.7 ± 3.0

TABLE 2. Percentage Differences of Pre and Post-operative Speech Scores Among Different Randall Class Patients for the Type of Procedure

Randall Class or Type of Procedure		Preoperative Speech Score	Postoperative Speech Score	Difference	*P**	Percentage Improvement
Overall	PR only	12.15 ± 0.95	7.32 ± 2.28	4.82	0.000	39.75%
	PR + PF	11.30 ± 1.83	3.76 ± 2.71	7.53	0.000	66.72%
Randall 1	PR only	12.07 ± 0.27	7.69 ± 1.54	4.38	0.000	36.29%
	PR + PF	10.50 ± 2.54	5.00 ± 3.49	5.50	0.000	52.38%
Randall 2	PR only	12.12 ± 1.07	7.16 ± 2.50	4.96	0.000	40.92%
	PR + PF	11.34 ± 1.68	3.46 ± 2.63	7.88	0.000	69.49%
Randall 3	PR only	12.50 ± 0.755	7.87 ± 1.64	4.62	0.001	37.04%
	PR + PF	12.25 ± 0.70	3.50 ± 1.06	8.75	0.000	71.43%

reported addition of primary pharyngeal flap for cleft palate repairs on the rationale that mesoderm in soft cleft palate is deficient; nevertheless, they were not sure whether addition of pharyngeal flap at the time of primary repair has any added advantage. Murthy et al[8] studied the speech outcome of patients in which late repair of the palate has been performed, and they concluded that moderate but definitive improvement occurs in such patients. Recently, pushback pharyngeal flap has been found to be safe and effective technique for velopharyngeal insufficiency.[19,20] All these studies show that there is need for some addition of tissue to primary cleft palate repair in late palate repair patients.

In the past, a few surgeons tried pharyngeal flap for wide cleft palate, but found it to increase the incidence of sleep apnea in small children.[22] The current study includes population of 6 years or older above, and it may be of the reasons for reduced chances of sleep apnea. Second, our technique of adjusting the pharyngeal flap to palate repair is somewhat unique in the sense that we utilize only 60% of the width of posterior pharyngeal wall rather than 80% or more as mentioned in literature.[23] This size is very effective in reducing the risk of obstructive sleep apnea in our patients. In addition, the way in which the pharyngeal flap is inset is very helpful as chances of contraction or dehiscence of the flap are very low. Second, because of the attachment of the muscles of soft palate with the pharyngeal flap, the flap acts like a dynamic structure as it moves with the soft palate musculature.

The authors have utilized the Pittsburgh Weighted Speech Score System[17] for speech assessment. Different studies have used subjective as well as objective assessment tools for speech assessment. Wattanawong et al[24] have carried out perceptual as well as instrumental assessment for comparison of pharyngeal flaps. Rochlin et al[20] also assessed the speech of patients, preoperatively and postoperatively by clinical instruments in addition to nasometry. Murthy et al[8] assessed the preoperative and postoperative speech in late palate repair patients by speech language pathologists who recorded the speech samples with a microphone. Keuning et al[25] studied the intrajudge reliability of perceptual rating of cleft palate speech before and after pharyngeal flap surgery and concluded that speech language pathologists do not guarantee a high intrajudge reliability of rating. The authors relied on subjective assessment as speech improvement is the primary objective of any surgery for cleft palate and velopharyngeal incompetence, and subjective methods can be relied on in such cases.

There are a few limitations to this study. It is the experience of a single surgeon from his 2 centers. Other surgeons may find different results depending on their experience and patient variability. Objective measurements such as nasometry or videofluoroscopy have not been carried out. Nevertheless, this study has a large number of patients in which different Randall types have been stratified to see the effect of addition of pharyngeal flap in different categories. In addition, the speech pathologist was blinded about the procedure, and so was the data analyst.

We propose that multicenter trials be carried out at different regions of the world to assess the suitability of pharyngeal flap at the time of primary palate repair. This would help in formulating the protocol for

TABLE 3. Elaborated Speech Score Values for Different Groups of Patients According to the Procedure (Palate Repair [PR Only] and Palate Repair With Pharyngeal Flap [PR + PF]) Among Different Subgroups of Randall's Type

		Preoperative Speech Score						Postoperative Speech Score					
Randall Class	No. Patients	Overall	Nasality	Nasal Air Emission	Phonation	Articulation	Facial Grimacing	Overall	Nasality	Nasal Air Emission	Phonation	Articulation	Facial Grimacing
Overall PR only	76	12.15	2.5	2.4	2.5	3.5	1.2	7.32	1.06	1.00	1.16	2.9	1.2
Overall PR + PF	63	11.30	2.3	2.3	2.4	3.3	1.0	3.76	0.4	0.4	0.3	2.2	0.46
Randall 1 PR only	13	12.07	2.5	2.4	2.2	3.5	1.4	7.69	1.1	1.0	1.1	3.2	1.2
Randall 1 PR + PF	12	10.50	2.2	2.1	2.3	3.0	0.9	5.00	0.7	0.9	0.5	2.6	0.3
Randall 2 PR only	55	12.12	2.4	2.3	2.6	3.3	1.5	7.16	1.0	1.8	1.3	2.8	0.26
Randall 2 PR + PF	43	11.34	2.3	2.4	2.4	3.2	1.0	3.46	0.3	0.2	0.1	2.3	0.5
Randall 3 PR only	8	12.5	2.6	2.4	2.7	3.7	1.1	7.87	1.1	1.2	1.1	3.2	1.27
Randall 3 PR + PF	8	12.25	2.5	2.3	2.6	3.8	1.05	3.50	0.2	0.3	0.3	2.4	0.3

management of young cleft palate patients who would benefit from such intervention, in reducing the number of surgeries and improvement of speech. In addition, the benefit of pharyngeal flap to reduce the duration of speech therapy sessions also needs to be explored.

CONCLUSIONS

In select group of patients who report late for palate repair, addition of pharyngeal flap along with the primary palate repair improves the speech outcome in all Randall groups.

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