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Investigation of the speech results of posterior pharyngeal wall augmentation with fat grafting for treatment of velopharyngeal insufficiency *



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ABSTRACT

Purpose: The purpose of this study was to evaluate the speech results of posterior pharyngeal wall augmentation (PPWA) with fat grafting both in the early and late postoperative period, and to clarify the impact of the procedure concomitant with speech therapy.

Materials and methods: This is a prospective case-control study. The study involved 87 cleft palate \pm cleft lip patients with velopharyngeal insufficiency (VPI) who has been treated with PPWA. Patients were separated into two groups according to age; the first group consisted of 49 pediatric participants between 6 and 12 years of age and the second group consisted of 38 adolescent participants between 13 and 18 years of age. Preoperative velopharyngeal function and articulation were compared postoperatively at the following time points: the 3rd month, 12th month, 18th month and 24th month. The velopharyngeal function was evaluated with regards to the velopharyngeal closure type and velopharyngeal closure amount, by using the pediatric flexible nasoendoscopy and the nasometer methods. In the nasometer evaluation, nasalance sores were measured by using nonsense syllables and meaningful sentences. The Ankara Articulation Test (AAT) (Ege et al., 2004) was used to detect compensatory articulation products secondary to VPI. Consonant production error types and frequencies were determined according the guidelines stated in the study of Hardin-Jones et al. (2009). These were Pharyngeal Fricatives - Posterior Nasal Fricatives/Stop Production, Glottal Stop Production, Middorsum Palatal Stop Production, Nasal Frictional Production, Posterior Nasal Frictional Production/Phoneme Specific Nasal Emission, use of Nasal Consonants for Oral Consonants, and Replacement of Trills. All the participants received concurrent speech therapy four times, twice in the post-operative period between 1 and 3 months and twice between 3 and 6 months.

Results: PPWA improved the speech performance from the 18th month to 24th month of the postoperative period. AAT assessment of the first group after 24 months comparing the post-PPWA with the preoperative data showed a highly significant decrease with regard to compensatory production errors

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and hypernasality; however, in the second group, the same comparison revealed a highly significant decrease in regard to the degree of hypernasality and a significant difference in terms of glottal articulation and pharyngealization of fricatives. A circular closure pattern was observed in 17 individuals with cleft palate at a rate of 70.6%.

Conclusion: PPWA with concurrent speech therapy is an acceptable surgical method to correct VPI and to improve speech performance.

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1. Introduction

One of the most important functional problems observed in individuals with cleft lip-palate is speech disorders. The main purpose of palate surgery is to achieve successful speech results without limiting the potential for maxillary growth (Witt and Kummer, 2009; Fisher and Sommerlad, 2011). Velopharyngeal insufficiency (VPI) refers to the non-convergence of the velum against the posterior pharyngeal wall during speech. The ability to close the velopharyngeal port can ultimately reduce abnormal resonance, hypnotic speech and intelligibility (Bishop et al., 2014). The surgical technique and surgical timing applied in VPI is quite important (Kummer, 2014a, 2014b, 2014c; Gart and Gosain, 2014). Determination of velopharyngeal closing pattern is especially important in the choice of surgical procedure for the treatment (Woo, 2012). Surgical techniques applied to correct VPI include pharyngeal flap, sphincter pharyngoplasty, Furlow's double opposing Z-plasty and posterior pharyngeal wall augmentation (Kummer et al., 2006; Gart and Gosain, 2014) If there is minimal insufficiency in soft palate motility and satisfactory lateral pharyngeal wall movement resulting in a minimal velopharyngeal gap, then PPWA is an ideal technique (Denny et al., 1993; Bishop et al., 2014). Currently, augmentation of the posterior pharyngeal wall using fat grafting is gaining popularity, as it is a less invasive alternative (Cantarella et al., 2011). Examination of the velopharyngeal closure pattern is quite important in terms of treatment planning (Croft et al., 1981; Sullivan et al., 2011). With the analyses done in the pre-operative period, identification of the closure type and planning of the surgical treatment to be chosen accordingly affect the results in a positive manner (Schuster et al., 2006).

Our hypothesis was that PPWA would quantitatively and qualitatively eliminate VPI and improve speech performance. In the studies conducted, it has been argued that there is a relationship between velopharyngeal dysfunction and speech disorders (Kummer, 2014a, 2014b, 2014c). In individuals with cleft palate, velopharyngeal function is affected by numerous symptoms that affect speech articulation/phonetics (Peterson-Falzone et al., 2010; Scarmagnani et al., 2015; Sweeney and Sell, 2008). During speech, as a result of oral and nasal cavities not being fully separated, mandatory and compensatory speech disorders may be observed. One of the most frequently seen disorders is the "glottal stop production" errors (Kulak Kayıkcı, 2015; Schuster et al., 2006, Albustanji et al., 2014; Esen Aydınlı et al., 2015). Improved articulatory placement through speech therapy may eliminate compensatory errors, improve velopharyngeal function, minimize perception of hypernasality, and improve speech intelligibility (Ghandour et al., 2013). The results provide important information for SLPs to evaluate and improve speech outcome in cleft palate patients.

The aim of this study was to evaluate velopharyngeal function and speech results before and after the application of PPWA surgery.

2. Materials and methods

In the present study, individuals who applied to Hacettepe University's Cleft Lip Palate ($CL \pm P$) councils for the first or a control evaluation were included. All the evaluations were carried out in the Department of Ear, Nose, and Throat and the Audiology and Speech Pathology Unit at Hacettepe University Hospital. This study was approved by the Ethics Committee of Hacettepe University (Approval Number: GO 14/588-29). All of the children's parents gave informed consent for participation, consistent with the Code of Ethics of the World Medical Association (Declaration of Helsinki).

In this study, a total of 87 individuals were included after they were diagnosed with VPI and applied PPWA, who were 6–12 years of age in the pediatric period (n = 49) or 13–18 years of age in the adolescent period (n = 38). Each individual's hospital records were analyzed and recorded. Inclusion criteria were as follows: having been diagnosed with cleft palate + cleft lip, decision taken by the $CL \pm P$ council members to apply the PPWA technique on the patient (plastic surgeon-speech language pathologist [SLP]orthodontist, otorhinolaryngologist), not having had primary surgery, not having fistulas on examination of the inner mouth, not having hearing loss, and not having a language and speech disorder other than speech disorders related to velopharyngeal dysfunction. Mental ability was age appropriate for all patients. On the evaluation day, the patients selected were without influenza or upper respiratory tract infection, which might affect the results of examinations. Cleft type was recorded in respect to the Veau classification.

2.1. Surgical technique

The procedure was performed under general anesthesia. The fat was harvested from the lower abdomen or inner thigh by liposuction under mild light pressure. A mouth gag was used to reveal the nasopharynx. The liposuccinate was centrifuged at 1200 g to obtain a 3-fold quantity. Fat graft was applied into the velum and the posterior and lateral nasopharyngeal walls using blunt tip injection cannulas. The study included 87 patients with VPI injected with 3.5–8 mL fat and soft palate under general anesthesia in the posterior, lateral pharyngeal walls. No morbidity of the donor site or injection site was observed. CPL The Council adopts the closure model when determining surgical technique.

2.2. Data collection

For evaluating velopharyngeal function and for instrumental evaluation methods, a pediatric flexible nasoendoscopy method (Croft et al., 1981) and nasometer (Watterson et al., 1999) were used. Nasometry is a computer-based tool that quantifies nasal air escape and allows comparison of the score against normative data. Acoustic signals were calculated by using a nasometer as synchronic (Kummer, 2014a, 2014b, 2014c) In this study, Nasometer II, Model 6450, (Kay Elemetrics Corp., Lincoln Park, NJ, USA)

Sentences with weighted and also sentences with nasal consonants and counts to 10 from 1, which include high intraoral pressure consonants. In the nasality evaluation, the average nasality scores were recorded and classified additionally for the analyses as 0-30% = normal, 30-40% = slight level of hypernasality, 40-60% = medium level hypernasality and >60\% = severe level of hypernasality (Kummer et al., 2006; Scarmagnani et al., 2015). In our study, nasometer evaluation was done at five different periods for each patient. For determining the closure status of the velopharyngeal area during speech, a flexible nasoendoscopic evaluation was used.

All procedures were done while the individuals were sitting by themselves or, if they were young, while they were in their mothers' arms. An ENT who was expert at his department performed the procedures. Images were recorded for evaluation in terms of velopharyngeal closure patterns. When the velopharyngeal area was arrived at with the endoscope, individuals were asked to repeat [pa, ta, ka, sa] syllables and to count from 1 to 10, which includes high intraoral pressure consonants. This practice lasted almost 10 min for every participant. During the practice, the process was recorded on video and the same SLP investigated twice.

Velopharyngeal closure type was determined by an experienced plastic surgeon, ENTs and SLPs as velopharyngeal closure types; 1 = coronal, 2 = circular, 3 = sagittal, and 4 = circular with the help of a Passavant ridge (Kummer and Lee, 1996; Scarmagnani et al., 2015).

Articulation assessment was conducted using a standardized Ankara Articulation Test (AAT) (Ege et al., 2004). The test was performed by SLPs in a silent room. Digital audio and video recordings were taken using a Sony Handycam HDR-CX11E (Sony Corporation, Tokyo, Japan) with a built-in microphone. During the evaluations, the outputs of individuals were recorded phonetically as much as possible. Evaluations were completed in approximately 25-40 min. Later, the recordings were listened to again, in a silent room, to check for the International Phonetic Alphabet (IPA) coding and symbols used for cleft-related errors and also were investigated with ASHA-IPA sound charts (Lenard, 2016). Consonant production error categories were presented under the main titles as suggested by Hardin-Jones et al. (2009). These were summarized as main titles as suggested by Brandt and Morris (1965) and presented as categories. These recordings had been previously edited by three SLPs. Every recording was analyzed live and with the audio recording.

2.3. Statistical analysis

The IBM SPSS Statistics 21 program (IBM Corporation, Armonk, NY, USA) was used in this study's statistical analysis. In the statistical evaluation of the findings, whether there is a difference between the groups in terms of numeric variables for the dependentpaired two-group difference was analyzed with the Wilcoxon test, and the group difference of more than two groups was analyzed with the Friedman test. In the independent samples, the Mann–Whitney U test was used to determine the different between two groups. In the analysis of the relationship between categorical variables, the chi-squared independence test was used. In the chi-square test, if the crosstab cell value related to categorical variables was 5 or higher in value, the Fisher test was used. In order to determine the amount and direction of the relationship between categorical variables, the Spearman correlation value was used. The significance value was accepted as 0.05.

3. Results

Within the scope of our study, the percentage of velopharyngeal closure type in 87 individuals with cleft palates have been analyzed through PFN and a 70.6% circular closure pattern has been observed. The type of palatal clefts included a veau 3 (%65.3) and veau 4 (%34.7).

Table 1 displays nasalance scores according to all speech samples in both groups. Nasality scores of/pi/and/numbers/are significantly higher than with the other speech samples (p < 0.05). When the groups are compared, the nasalance scores were detected significantly lower at the 18th and 24th months (p < 0.05) in the first group. The nasalance scores of all speech samples were lower at the 24th month (p < 0.05). When the two groups were compared, an earlier decrease in nasalance scores were observed in the pediatric group (p = 0.039).

Table 2 shows the comparison of consonant production error categories in the five different time points in both groups. When the two groups are compared, a significant difference was found in the glottal stop production errors and middorsum palatal stop production errors between the 1st and the 2nd groups in the preoperative period (Mann–Whitney U = 11.00, p = 0.018 < α = 0.05, Mann–Whitney U = 11.08, p = $0.000 < \alpha = 0.05$). In addition, according to the preoperative period, number of consonant output error was higher in group I than in the other group (p < 0.05). When the two groups were compared postoperatively, there was a highly significant decrease found in all parameters on comparing the second group, which revealed that there was a nonsignificant difference in regard to all parameters (glottal stop, pharyngeal fricative, nasal consonant use for oral consonants, replacement of thrills, middorsum palatal stop) according to five different time points (p < 0.05). It was determined that the number of glottal stop production errors had decreased (Mann-Whitney U = 31.00, $p = 0.047 < \alpha = 0.05$). When the average sequence values were evaluated, it was seen that the glottal stop consonant error production difference between the first group (8.60) and the second group were close in value.

In the group analysis, in the first group, glottal stop consonant production was decreased just after the surgery and middorsum palatal stop consonant error was decreased in the period of 18th to the 24th month (p = 0.000). In the second group, glottal stop consonant production was decreased in the period of the 18th to the 24th month (p = 0.047).

4. Discussion

Augmentation of the posterior pharyngeal wall can be a less invasive alternative and can be achieved by fat grafting (Cantarella et al., 2011). When a palatal procedure is indicated, surgical approaches are tailored to address each individual's pattern of velopharyngeal closure deficiency. Therefore, the selection of suitable candidates according to closure pattern for PPWA, is important to keep in mind. Identification of the velopharyngeal closure pattern is quite important in terms of treatment planning (Croft et al., 1981). Planning changes greatly in accordance with the result to be obtained. In the literature, the coronal pattern is the most frequently seen closure pattern, the circular pattern is the second most frequently seen pattern, and the sagittal pattern is the least frequently seen pattern (Rowe and D'Antonio, 2005; Tieu et al., 2012). Within the scope of our study, the percentage of velopharyngeal closure type in 87 individuals with cleft palates was analyzed through PFN and a 70.6% circular closure pattern was

Table '	1
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Nasalance scores according to the recording times for speech samples [/pa/,/ta/,/ka/,/sa/,/pi/] syllables, counting from 1 to 10 in both groups	Nasalance scores accordin	g to the recording times for	speech samples [/pa/./ta/.	/ka/./sa/./pi/l svllables, coun	ting from 1 to 10 in both groups.
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	Recording times	Speech samples				Counting numbers		
	(Median + IQR)	/pa/	/pi/	/ta/	/sa/	/ka/	from 1 to 10	
Pre op	Group I	36.00 ± 12.00	33.00 ± 11.25	31.50 ± 13.50	31.00 ± 12.00	29.00 ± 14.50	56.00 ± 10.00	
	Group II	52.00 ± 18.25	63.50 ± 11.25	38.50 ± 11.50	35.00 ± 10.00	48.00 ± 11.50	45.00 ± 11.25	
Post op	Group I	62.00 ± 17.00	55.00 ± 16.75	49.50 ± 14.00	51.00 ± 16.75	46.00 ± 18.25*S	50.00 ± 11.25	
3rd M.	Group II	68.00 ± 14.00	45.00 ± 11.50	40.50 ± 11.50	27.00 ± 11.50	44.00 ± 14.25	41.00 ± 11.25	
Post op	Group I	39.00 ± 10.00	35.00 ± 10.00	33.00 ± 14.50	35.00 ± 10.00	31.00 ± 14.25	51.50 ± 11.50	
12th M.	Group II	52.00 ± 10.25	35.00 ± 10.50	36.50 ± 11.50	30.50 ± 10.25	45.00 ± 12.00	50.00 ± 11.25	
Post op	Group I	40.00 ± 12.00	31.00 ± 11.50	27.00 ± 11.50	29.00 ± 15.75	25.00 ± 14.25	45.00 ± 11.25	
18th M.	Group II	47.50 ± 14.00	34.00 ± 11.50	33.00 ± 11.50	26.00 ± 10.00	45.00 ± 17.00	52.00 ± 11.25	
Post op	Group I	34.00 ± 14.00	36.00 ± 12.50	36.00 ± 10.25	36.00 ± 14.00	32.50 ± 13.50	43.50 ± 10.25*S	
24th M.	Group II	27.00 ± 11.50* S	30.00 ± 11.50	20.00 ± 11.50	30.50 ± 10.25	40.00 ± 11.50	45.00 ± 11.25	

IQR, interquartile range; M, median; S, significant; Pre op, preoperative; Post op, postoperative.

Median and interquartile range of nasality scores according to the recording times for speech samples are shown. In all speech samples, nasality scores in preoperative period had higher values, and the difference was statistically important after the postoperative period, especially the sample of/pi/and counting numbers from 1 to 10. * Significant at p < 0.05 (p = 0.039).

Ta	hla	2

Comparison of consonant production error categories recordings taken at the five different time points in both groups.

Consonant production error Categories recordings taken in the five different time points in both groups			Mann–Whitney U	Median + IQR	р
Pre op	Glottal stop	Pediatric period	11.40	11.00 ± 31.00	0.018*
		Adolescent period	5.57		
Post op (month: 18th–24th)	Glottal stop	Pediatric period	8.60		.047*
		Adolescent period	9.75		
Pre op	Pharyngeal fricative	Pediatric period	7.50	20.00 ± 20.50	.141
		Adolescent period	11.14		
Post op (month: 18th–24th)	Pharyngeal fricative	Pediatric period	7.55		.146
		Adolescent period	11.07		
Pre-op	Nasal consonant	Pediatric period	8.90	34.00 ± 22.00	.919
	Use for oral consonants	Adolescent period	9.14		
Post op (18th–24th)	Nasal consonant	Pediatric period	7.70		.177
	Use for oral consonants	Adolescent period	10.86		
Pre-op	Replacement of trills	Pediatric period	10.40	21.00 ± 24.50	.144
		Adolescent period	7.00		
Post op (month: 18th–24th)	Replacement of trills	Pediatric period	10.05		.121
		Adolescent period	7.50		
Pre op	Middorsum palatal stop	Pediatric period	12.50	11.00 ± 19.00	.000*
		Adolescent period	4.00		
Post op (month: 18th–24th)	Middorsum palatal stop	Pediatric period	10.60		.102
		Adolescent period	6.71		

Pre op, preoperative; Post op, postoperative; IQR, interquartile range; M, median; S, significant.

Average values are given.

*p < 0.05 (significant at p < 0.05).

observed. It is considered that this value will have a positive effect on the results in terms of the identification of the circular closure type in the population with cleft palates and the planning of the PPWA surgical treatment to be chosen in accordance with individuals with cleft palates.

In the literature; few previous reports exist regarding the use of PPWA in individuals with VPI when speech performance is considered in participants with VPI. In Lypka et al.'s study (Lypka et al., 2010), 111 individuals who underwent posterior pharyngeal augmentation for the treatment of VPI were reviewed retrospectively in terms of age at the time of operation, type of implant used, duration, and speech performance. Speech performance was analyzed by the nasoendoscopy and four-point grading scale. As a result of the study, it was stated that PPWA is a safe and effective treatment for patients with VPI. Implants are well tolerated and speech is substantially improved. In Cao et al.'s study (Cao et al., 2013) in 11 individuals with cleft palates between the ages of 5 and 26 years who were diagnosed with VPI, a positive development has been observed in terms of speech articulation/phonetics and pronunciation guality in the evaluation made 40 months after PPWA was applied. As a result of the study, it was stated that in VPI treatment, PPWA has been an alternative invasive method

compared to surgical techniques of palatoplasty and pharyngoplasty (Sader et al., 2015; Pet et al., 2015). The impact of the patient populations on the speech results achieved with autologous fat grafting is underscored by comparing the 2011 and 2013 studies of Filip et al. (Bishop et al., 2014).

In the present study, when speech performance is considered, it was observed in the postoperative period that the number of glottal stop production errors and the number of middorsum palatal stop production errors decreased after the 18th month of the postoperative period. The present study differs from other studies presented above by the speech evaluation method, since the standardized articulation test was not only used at the beginning but was also repeated in the follow-up period. In addition, it included evaluating the compensatory error production types. This provided a more reliable means of exploring the effect of PPWA surgery on speech performance and determining the exact time period for speech improvement in two distinct age groups. The aim of dividing participants into two groups was to evaluate articulation performance in more homogeneous groups.

In the literature, there are a limited number of studies on the velopharyngeal function status in terms of nasality comparison after PPWA surgery (Dejonckere and Van Wijngaarden, 2000; Lau et al., 2013; Lauchter et al., 2010; Wójcicki and Wójcicka, 2011; Bishop et al., 2014; Gray et al., 1999). In Dejonckere and Van Wijngaarden's study (Dejonckere and Van Wijngaarden, 2000), it was observed that in 17 individuals with cleft palates (4-24 years old; mean, 9.7 years) who had been diagnosed with mild VPI between 1996 and 1999, there was a decrease of 30% in nasality scores after the 6th month after the PPWA. The authors stated that autologous fat seems to be an excellent alternative in this indication and that nasometry allows a precise quantitative assessment of functional velopharyngeal surgery. In Wójcicki et al.'s study (Wójcicki and Wójcicka, 2011), it was seen that the hypernasality in the 6th month after PPWA decreased from 48% to 33%. In Gray et al.'s study (1999), additionally, a correlation was found between age and nasometry improvement after PPWA. They found that younger patients did better. In the literature, there are a limited number of studies on velopharyngeal function status in terms of nasality comparison after PPWA surgery (Dejonckere and Van Wijngaarden, 2000; Lau et al., 2013; Lauchter et al., 2010). Leuchter et al., in 18 patients with mild velopharyngeal insufficiency, examined the fat grafting results between 8 and 53 years of age. The hypernasal speech grade was evaluated preoperatively and postoperatively at 2 weeks, 2 months, 6 months, and 1 year. The mean value of the nasalance scores after PPWA was 37% preoperatively and 23% postoperatively (p = 0.015). Hypernasality decreased postoperatively in all patients (Leuchter, 2009). In all three patients, nausea improved significantly. After the fat grafting procedure, hypernasality completely resolved (Lauchter et al., 2010). Although the mean age of the patients was marginally younger than with Leutcher et al., it was unlikely to account for the different results between these studies (Bishop et al., 2014).

Piotet et al. (2015) evaluated fat grafting results in 22 patients with velopharyngeal insufficiency associated with palate clefts between 2004 and 2005 and evaluated short-term (within 2 months) and long-term efficacy (24 months). Results of post-operative nasalance scores were statistically significant and remained stable in most patients until the end of follow-up (median 42 months), and autologous fat injection was a simple procedure for treatment failure. Patients with cleft palate had good long-term outcomes and few complications (Leboulanger et al., 2011). In a prospective study, Cantarella et al. (2011) examined the effectiveness of injecting the posterior and lateral pharyngeal walls and the velum in patients with a VP closure gap of less than 50%. They achieved a reduction of the velopharyngeal sphincter in all of their patients and a significant improvement in hypernasality and nasal air escape (Bishop et al., 2014).

Most individuals with cleft lip and palate have articulation problems because of compensatory articulation disorders from velopharyngeal insufficiency. Demark et al.'s analysis of the data indicate that, as a group, these subjects with cleft palate were retarded in articulation skills (1979). In light of such a relationship between velopharyngeal insufficiency and articulation errors, one would expect to obtain relatively high correlation coefficients between these two variables (Branth and Morris, 1965). Ysunza-Rivera et al. (1991) studied 31 patients with surgically repaired cleft palates who had VPI and compensatory articulatory defects. All patients were subjected to objective methods of multiple incidences before and after speech therapy in order to correct the compensatory articulation. The movement proportions of the pharyngeal velum structures increased significantly after correction of the compensatory articulation.

As a result of the present study, it has been seen that the PPWA application reduces hypernasality. In our nasalance score results, nine patients had had hypernasality. After the operation, two patients were found to be free from nasality, eight had slight nasality, and four had medium nasality. Before the operation, all patients had an abnormal nasalance score, with the mean value above 63%. After the operation, only three patients had values ranking between 25% and 33%, and they were found to have moderate hypernasality, whereas the mean value decreased to 24%. A total of 49 patients achieved full recovery (74%) in the pediatric group, and the remaining four patients had improved recovery (26%) in the adolescent group. These latter patients gualified for further speech therapy. In our study, it is seen that the nasalance scores of each speech sample taken from individuals with cleft palates who have been diagnosed with VPI in the preoperative period is higher compared to the nasalance scores in the postoperative period. In general, when the average values of the tables are analyzed, it can be seen that the nasalance scores of the/pi/and/numbers/speech samples among the speech samples in the preoperative period are higher than the other samples. It has been seen that the decreasing of the nasalance score values was more prominent in the pediatric group in the 18th month, whereas the same decreasing trend was seen in the adolescent group by the 24th month.

5. Conclusion

To summarize, this study is one of the most comprehensive studies to examine the effect of PPWA surgery not only by evaluating velopharyngeal function but also speech articulation with a follow-up period. On the basis of clinical observation of 87 patients during a preoperative to 24-month (mean, 24.5 months), it is suggested that PPWA is a promising method for treating velopharyngeal insufficiency. It is considered, for future studies, grouping the number of consonant error production categories and the number of individuals who receive PPWA after being diagnosed with VPI in terms of undergoing or not undergoing operation and early versus late operation time, and analyzing the operation's effect on articulation skills might give beneficial results.

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