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The incidence of velopharyngeal insufficiency and oronasal fistula after primary palatal surgery with Sommerlad intravelar veloplasty: A retrospective study in Isfahan Cleft Care Team

Dorsa Mapar, Fatemeh Khanlar, Saba Sadeghi, Hossein Abdali, Mehrdad Memarzadeh, Heydar Ali Davari, Fatemeh Derakhshandeh*

Craniofacial & Cleft Research Center, Isfahan University of Medical Sciences, Isfahan, Iran

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ABSTRACT

Objectives: This study was designed to determine the incidence of velopharyngeal insufficiency (VPI), oronasal fistula development and facial grimace in patients seen by Isfahan Cleft Care Team (ICCT) after primary Sommerlad intravelar veloplasty (SIVV). Furthermore the association of gender, cleft type and age at primary surgery with the incidence of hypernasality and fistula is determined.

Methods: A group of 40 patients with history of cleft palate with or without cleft lip were identified from the records of ICCT between 2011 and 2014. The main outcome measures were the incidence of hypernasality and fistula after primary palate repair with SIVV. Speech recordings were analyzed by consensus by two speech therapists according to the Cleft Audit Protocol for Speech- Augmented (CAPS-A), (Kappa = 82.4). Deciding whether or not to have a fistula was based on the oral examination videos.

Results: Severe and moderate hypernasality was observed in 42.5% of patients. Normal resonance and mild/borderline hypernasality was observed in 37.5% and 20% of patients, respectively. The frequency of fistulas was 7.5%. There was a significant association between hypernasality with cleft type and the age at primary surgery ($p < 0.05$).

Conclusion: Significant progress has been made in the outcomes of the primary palate surgeries with the SIVV technique compared to the previous study in the ICCT.

1. Introduction

Primary palatal surgery is the first physical intervention to repair the cleft palate and has a determining role in the quality of speech outcomes [1,2]. The aim of this surgery is to provide a proper structure between the mouth and nose, improve the nutritional state and hearing and decrease respiratory infections [3,4]. The main objective of this surgery is to create a healthy structure for normal speech production, direct oral airflow, and a balance in resonance with minimal effects on the facial growth [2,3].

There are different techniques for primary palatal surgery, including the Von-Longenbeck, Furlow Z-plasty and Sommerlad intravelar veloplasty (SIVV). This paper describes the results of primary palate repair with the last one (SIVV). In this technique, which is performed under the operating microscope, the levator is inserted into the margin of the cleft more anteriorly than normal and, at operation, is retrodisplaced to the middle of the velum or, usually, even further back, to increase the functional length of the palate and improve its function. In fact, this surgery is cleft palate repair with minimal hard palate dissection and radical muscle reconstruction [2,3,5].

Different studies use different criteria to report the results of speech following primary palatal surgery. Among these criteria, hypernasality is widely used as the most common postoperative resonance disorder and to predict velopharyngeal insufficiency (VPI)[1]. In addition, there is a known interaction between VP function and fistula that the presence of oronasal fistula after primary palatal surgery may increase hypernasality [1].

Many studies have investigated the most important speech findings following primary palatal surgery, i.e. oronasal fistula and hypernasality. Ha et al. conducted a study to report speech outcomes following primary palatal surgery. They studied 292 non-syndromic patients that underwent different surgical techniques, and reported hypernasality and oronasal fistula in 20.8% and 7.9% of the patients, respectively [6]. Mahoney et al. reported rates of 0.3% and 13.8% for oronasal fistula

* Corresponding author.
E-mail address: derakhshandeh.fateme@gmail.com (F. Derakhshandeh).

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and the requirement for secondary surgery following different surgeries for palate repair [7]. Aslam et al. reported a frequency of 5.6% for oronasal fistula following complete cleft palate repair [8].

Hypernasality occurs in approximately 70–82% of patients following primary palatoplasty in ICCT in 2004–2010 [9–11].

Significant correlation between hypernasality and the type of cleft [6,9,10], hypernasality and age at primary surgery [9,10,12] and fistula and the cleft type was observed [7].

Considering the high incidence of VP insufficiency in ICCT's studies [9–11] as well as the benefits of the SIVV which is improved VP function [2,6,13–17], the Isfahan cleft team started using this technique since 2011. So far, no study has evaluated the speech outcomes of this technique. Therefore, the present study was conducted to evaluate the incidence of oronasal fistula and hypernasality following primary palatal surgery using intravelar veloplasty. In addition, the association of hypernasality and oronasal fistula with gender, type of cleft, and age at primary surgery was investigated.

2. Materials and methods

2.1. Participant

Medical files of 2200 patients were reviewed and 104 files of the patients that underwent SIVV as a primary palate repair technique were selected. Sixty four of 104 files were excluded because of age below three years at assessment time, syndromes or other congenital anomalies except cleft palate, cognitive disorders, neurological diseases, and sensorineural hearing loss as well as inappropriate quality of voices and videos. Therefore, 40 files were included in the final analysis(Table 1).

According to the age at primary surgery, the patients were divided into three groups: those under 12 months of age, 12–18 months, and after 18 months. According to the Veau system, the cleft type was categorized into soft cleft palate, hard/soft palate cleft, unilateral cleft lip and palate, and bilateral cleft lip and palate [12]. Also A subgroup of patients with submucous cleft was considered. The Cleft Palate Clinic archive was used to evaluate the children's speech samples and oral examination videos.

2.2. Recording/assessments procedure

In Isfahan Cleft Clinic, videos and voice samples are routinely recorded by the Sony handycam (HDR-PJ410) and a Sony sound recorder (ICD-SX2000) placed about 30 cm from the patient's mouth in a quiet room with natural light [18]. Both the audio and video recordings were transferred to an Acer lap top for listening and rating. Two speech-language pathologists (one expert in cleft palate and one trained researcher) scored all of the samples for hypernasality independently (kappa = 82.4%). According to the CAPS-A, the severity of hypernasality is scored as follows: absent (0), borderline (1), mild (2), moderate (3), severe (4).

The presence of oronasal fistula were rated after watching the oral examination videos by the main researcher. All the posterior fistula (in hard and soft palate) were considered. Fistulas above 5 mm were included in this study.

In this study, the compensatory articulation errors were not investigated.

Table 1
Flowchart of inclusion and exclusion of patients(N = 40).

		NO.
Exclusion patients	Children under 3 years	167
	Surgery with other technique except SIVV	218
	syndromes or other congenital anomalies	23
	Secondary Surgery or surgery in other hospitals or	1654
	without surgery	62
	Insufficient follow ups and medical reports	36
	Lack of speech assessments/Failure to refer to speech evaluation	
Inclusion patients		40
Total		2200

2.3. Speech sample

Patients were asked to repeat the Persian sentences with high pressure consonants developed according to the Universal Parameters guidelines [19] and read a poem as connected speech based on the CAPS-A.

2.4. Data analysis

The Statistical Package for Social Sciences (SPSS) version 22 was used for data analysis. At first, the frequency of hypernasality and oronasal fistula was calculated; then, the correlation of these two variables with gender, age at primary surgery and cleft type was analyzed using non-parametric tests (chi-squared and Spearman correlation).

2.5. Surgeons

The surgeons were the same as the last study [11]. They had at least 15 years of palate surgery in other kinds of technique but they had experienced in SIVV technique between 3–5 years.

3. Results

Forty files (16 boys and 24 girls) were included in the analysis. The demographic and clinical characteristics of the subjects are presented in Table 2.

The median age at primary surgery, after excluding four outlier data, was 13.5 months (range: 6 months to 6 years, mean = 19.58 ± 2.88 months).

According to the results, 37.5% (n = 15) had a normal VP function (hypernasality severity = 0). The frequency of borderline, mild, moderate, and severe hypernasality is shown in Fig. 1. The patients who required secondary surgery due to moderate and severe hypernasality were 42.5% (n = 17), while 57.5% (n = 23) did not require further surgical interventions due to normal resonance or borderline/mild hypernasality.

Although the frequency of hypernasality was higher in boys versus girls (75% vs. 54.2%), this difference was not statistically significant (chi-squared test, p = 0.42). The percentage of subjects with normal resonance was 45.8% in girls and 25%in boys.

A significant correlation was found between the cleft type and hypernasality (p = 0.01). According to Table 3, the frequency of hypernasality was higher in patients suffering from submucous cleft and bilateral cleft lip and palate.

According to Table 4, the frequency of hypernasality increased significantly with an increase in age at primary surgery (p = 0.04). Oronasal fistula was seen in 7.5% of the patients (Fig. 2).

12.5% of boys (2 out of 16) and 4.1% of the girls (1 out of 24) had

Table 2
Gender and type of cleft in population (n = 40).

		No.	%
gender	male	16	40
	female	24	60
Type of cleft	soft cleft palate	7	17.5
	soft and hard cleft palate	9	22.5
	unilateral cleft lip and palate	9	22.5
	bilateral cleft lip and palate	9	22.5
	submucous cleft palate	6	15

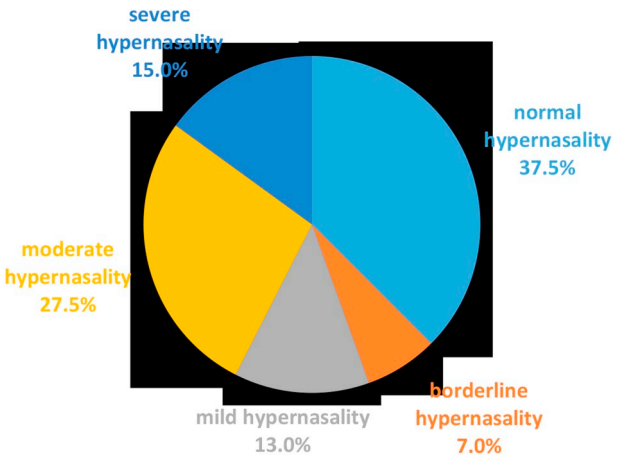

Fig. 1. Prevalence of hypernasality in population.

oronasal fistula but the differences were not significant (p = 0.38). Incidence of oronasal fistula was more common in bilateral cleft lip and palate compared to other types of cleft (Table 5), but was not statistically significant (p = 0.41).

No fistula was seen in individuals who were operated after 18 months while 2.5% (n = 1) and 5% (n = 2) patients in the age groups prior 12 months and 12–18 months had oronasal fistula (Table 4). There was no significant difference between age groups at primary surgery in this regard (p = 0.36).

4. Discussion

In this study, medical files of 40 patients who underwent primary palatal surgery using SIVV were reviewed and showed a frequency of 62.5% for hypernasality and 7.5% for oronasal fistula. Of these 40 subjects, 57.5% did not need to further surgery based on normal resonance, borderline and mild hypernasality, and 42.5% required to secondary surgery (with moderate and severe hypernasality).

The different rates of requiring secondary surgery was reported in different centers [2,6,9–11,20,21]. In general, it is difficult to compare the need for secondary surgery between different centers, because it depends on different factors such as the inclusion and exclusion criteria, the threshold of the cleft team, the parents' and/or children's wishes, and follow-up duration [5]. Moreover, there is no gold standard for comparison [22].

Regarding to high incidence of VPI based on the last studies conducted by ICCT [9–11], the team changed the technique of palate repair to SIVV from 2011. The results of the recent study showed a significant reduction in hypernasality and fistula rate due to the new technique, in which levator muscle correction is done [23]. One of the advantages of the SIVV is repair of the levator muscle, which is the main muscle for

Table 4
Rates of hypernasality and oronasal fistula according to timing of palate repair.

Degree of hypernasality	12 < months		12–18 months		> 18 months	
	(n = 17)		(n = 13)		(n = 10)	
	No.	%.	No.	%.	No.	%.
Normal	9	52.9	4	30.8	2	20
Borderline	1	5.9	1	7.7	1	10
Mild	2	11.8	1	7.7	2	20
Moderate	4	23.5	5	38.5	2	20
Severe	1	5.9	2	15.4	3	30
oronasal fistula	1	2.5	2	5	0	0

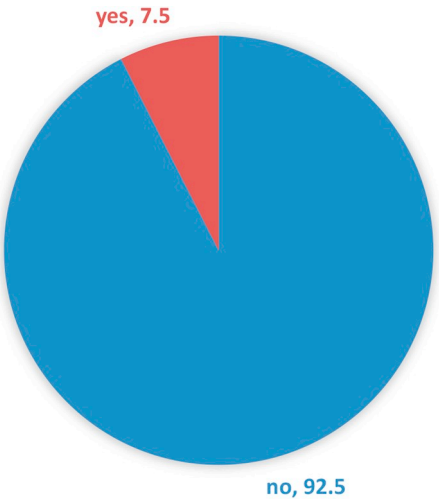

Fig. 2. Prevalence of fistula in population.

elevating the soft palate. Another advantage of the new technique is the use of a microscope for muscle correction. The microscope has a high quality, variable magnification, and acceptable illumination. Moreover, since it provides a direct image during surgery, it is more reliable than magnifiers and headlamps. Furthermore using a microscope provides the possibility of changing the viewing angle of binocular cameras to facilitate the surgery [24]. The last advantage of the new technique is palate repairing without any mucopriosteal falp elevation or lateral incisions that decreases the rate of oronasal fistula and scar formation and improves the palatal movement and speech outcomes [5].

Although speech outcomes of this study were improved dramatically in compare to the last researches conducting by ICCT, but the percentage of the patients who required secondary surgery is still high

Table 3
Rates of hypernasality according to type of cleft palate.

Degree of hypernasality	Veau I		Veau II		Veau III		Veau IV		SMCP	
	(n = 7)		(n = 9)		(n = 9)		(n = 9)		(n = 6)	
	No.	%.	No.	%.	No.	%.	No.	%.	No.	%.
Normal	5	71.4	4	44.5	3	33.3	2	22.2	1	16.7
Borderline	0	0	2	22.2	1	11.1	0	0	0	0
Mild	1	14.3	0	0	1	11.1	1	11.1	2	33.3
Moderate	0	0	2	22.2	3	33.3	5	55.6	1	16.7
Severe	1	14.3	1	11.1	1	11.1	1	11.1	2	33.3
	7		9		3		9		6	

Table 5
Occurrence of oronasal fistula in different types of cleft palate.

oronasal fistula	Veau I		Veau II		Veau III		Veau IV		SMCP	
	(n = 7)		(n = 9)		(n = 9)		(n = 9)		(n = 6)	
	No.	%.	No.	%.	No.	%.	No.	%.	No.	%.
fistula	0	0	0	0	1	11.1	2	22.2	0	0
no fistula	7	100	9	100	8	88.8	7	77.8	6	100

compared to the Sommerlad's study (42.5% vs. 10.2%). The reason for this difference may be that Sommerlad reported the results of operations in three 5-year periods from 1978 to 1992, during which the need for secondary surgery decreased from 10.2% to 4.6%. Therefore, it can be concluded that the surgeon's experience has a significant role in speech outcomes [5].

Analysis of the results according to the cleft type showed that the extent of the cleft is an effective factor in postoperative hypernasality. In the other words, the need for secondary surgery in soft cleft palate, hard/soft cleft palate, unilateral cleft lip/palate, submucous cleft palate and bilateral cleft lip/palate increases accordingly. Generally, the results of primary surgery, both with or without SIVV, were not favorable for submucous cleft palate and bilateral cleft lip/palate. The Spearman correlation coefficient also showed a direct and significant association between these two variables ($p = 0.02$, $r = 0.36$), which is similar to the results of several studies [6,10,12,25].

In this study, early palatal repair resulted in better VP function ($p = 0.06$, $r = 0.3$). This finding is confirmed with the results of the studies conducted by Hardin Jones and David Jones, Derakhshandeh et al., and Davari et al. [9,10,12].

Few studies have evaluated the correlation of sex and hypernasality [7,9,11,26] and reported different results. In the current study, despite the higher severity of hypernasality in boys than girls, no significant correlation was seen between sex and hypernasality ($p = 0.42$). The reason for the higher prevalence of hypernasality in boys may be higher prevalence of bilateral cleft lip and palate in this sex, and the existence of a correlation between the extent of the cleft and hypernasality. However, genetic, anatomical, and speech growth differences may also contribute to this difference [27].

The frequency of oronasal fistula varies from 0 to 60% [4,26,28,29]. A possible reason for this variation may be the surgeon's expertise [29–31]. Sommerlad reported a frequency of 15% for oronasal fistula after SIVV and stated that excluding bilateral cleft cases reduces the percentage of oronasal fistula from 15% to 12% [5]. In addition, the extent of the cleft also affects the rate of fistula formation [28]. In this study, the frequency of fistula formation was 7.5%. Although the fistula rate was increased with increasing the extent of the cleft and this finding was confirmed in the other studies [26,30,32], but the statistical analysis of this study showed no significant difference between cleft types ($p = 0.41$). In another study that used the same technique, oronasal fistula was seen in 3%, 3%, and 10% of the subjects with soft cleft palate, unilateral cleft lip/palate, and bilateral cleft lip/palate, respectively [32].

5. Limitation

A limitation of this study is that patients with normal resonance or borderline/mild hypernasality had weak participation in follow-up procedure in ICCT and this factor may resulted to more incidence of VPI and fistula in compare to when all the patients returned to team.

6. Conclusion

The Sommerlad intravelar veloplasty technique has been widely used by the Isfahan cleft palate team for the past seven years, and the occurrence of VPI has decreased about 30% over this period. We hope that with increasing the use of this technique in ICCT, the speech outcomes of primary palate repair will improve in the future as the surgeons gain more experience.

Conflicts of interest

There is no conflict of interest for doing this research.

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