

## Evaluation of outcome of Palatoplasty in patients with Isolated Cleft Palate Defect: A Retrospective Observational study.

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### ABSTRACT

**Objective:** To evaluate the outcome of palatoplasty in terms of closure of velopharyngeal gap and correction of speech in patients with isolated cleft palate.

**Study Design:** Retrospective observational study.

**Place and Duration:** ENT Department, Capital Hospital PGMI, Islamabad, Pakistan, from 1<sup>st</sup> January 2016 to 31<sup>st</sup> December 2017.

**Methodology:** Medical record of patients with isolated cleft palate having undergone palatoplasty from both genders aged 1-12 years were reviewed for demographic and clinical information. Data collected including age, gender, cleft type, age at palatoplasty, type of palatoplasty, post-palatoplasty velopharyngeal gap and pre and 6 months post-operative speech.

**Results:** Among total of 32 patients, good repair with palatoplasty was achieved in 68.8% cases and 31.3% were left with large velopharyngeal gap with significantly better repair results for secondary cleft compared to primary with  $p=0.002$ . In the  $\leq 1$  year age group post-operative velopharyngeal gap was normal while in the 1 to 4 year age group out of 26, 6 had large and 4 had moderate velopharyngeal gap ( $p=0.012$ ). Results also revealed significant ( $p=0.01$ ) improvement in speech results compared to pre-operative speech with significantly better results ( $p=0.007$ ) for secondary cleft.

**Conclusion:** Age at palatoplasty has significant association with velopharyngeal gap/ insufficiency ( $p=0.012$ ), while it has no significant association with speech results ( $p=0.415$ ) especially when comparing  $<1$  year and 1-4 years age group.

**Keywords:** Cleft palate, Palatoplasty, Age, Reconstruction, Langenbeck procedure, Rhinolalia, Velopharyngeal gap, V-Y pushback.

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### INTRODUCTION

Cleft palate (CP) is a rare developmental anomaly with failure of union of the bony and/or soft tissue elements of roof of oral cavity during the process of fetal development. Cleft palate has

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an incidence of 1.3 to 25.3 per 10,000 population. Of these 50% are non-syndromic clefts<sup>1</sup>. Local prevalence of cleft palate is interestingly very high (42.5%)<sup>2</sup>.

Palatal clefts can result in inability of the child to properly close the velopharyngeal sphincter i.e. velo-pharyngeal insufficiency (VPI). This can in turn, result in a number of issues including hyper-nasality and nasal regurgitation. Children with cleft palate can also face difficulties in feeding, dentition, hearing, speech and language development as well as compromised speech intelligibility<sup>3</sup>. Since these young children have to develop communication skills, surgical closure of the palatal defect to obtain a normal or near normal velopharyngeal mechanism for normal speech production is of utmost importance. A number of surgical interventions are in use including von Langenbeck procedure, V-Y pushback technique, Intra-velar veloplasty, Furlow's technique and Bardach & Salyer's Two-Flap palatoplasty<sup>4</sup>.

With no universal agreement on the most suitable surgical procedure<sup>4</sup>, further research is warranted<sup>5</sup>. A number of factors are incriminated to affect the outcome of palatal surgery which include different types and extent of clefts, level of early feeding and growth of baby, socioeconomic status, hearing status, stimulation level at home, some surgeon factors, age at time surgery, poor use of obturators, follow-up<sup>6</sup>, delayed presentation, and lack of involving allied professionals like speech language pathologists and orthodontists<sup>2</sup>. Hence, Tomas highlighted the

need of studies and widely varying claims of results of different procedures and recommended management with a multidisciplinary team<sup>4</sup>. Jajja et al. reported that median age of primary surgical repair was 13 months<sup>2</sup>.

In Pakistan the prevalence of cleft palate is quite high<sup>2</sup>, along with delayed presentation in tertiary care hospitals, where surgical intervention facility is available. Therefore, the surgical intervention in our cultural setup is delayed and might result in more cases with velopharyngeal insufficiency and poor speech compared to cases who receive early intervention. Also there is dearth of local literature on the subject, deficient knowledge regarding the pathology and its implications as well as need of early intervention. Hence, this needs to be studied, since it can affect the child's optimum communicational development.

Therefore the current study was conducted, with the hypothesis that delayed presentation between 1-4 years of age results in velopharyngeal gap/ insufficiency and poor speech results. This study has significant importance since it will add to local literature and highlight the need of timely intervention and also act as a statistical resource for future studies with different surgical strategies, which is the need of the hour. Hence this study was carried out with the objective to evaluate the outcome of palatoplasty to repair the defect and speech abnormalities in patients with isolated cleft palate.

#### METHODOLOGY

This retrospective observational study, reviewed charts of cases of isolated cleft palate who underwent palatoplasty at ENT Department, Capital Hospital, Postgraduate Medical Institute (PGMI), Islamabad over a period of 2 years from 1<sup>st</sup> January 2016 to 31<sup>st</sup> December 2017. Study included both genders, aged 1 to 12 years with isolated cleft palate operated at the facility with 6 months post-operative follow-up recorded in the ENT outdoor surgery follow up register. The Charts and record of follow up of these cases were reviewed by the researcher for demographic and clinical information including gender, cleft type, pre-operative speech, age at palatoplasty, type of palatoplasty, post-operative velopharyngeal (VP) gap and post-operative speech at 6 months. Cases with missing data and those in which VP gap and post-operative speech was not recorded at 6 months follow up were excluded from the study.

Pre-Intervention assessment comprised of history and ENT examination by an otolaryngologist, and speech assessment by a speech therapist. These included measurement of perceptual speech analysis and nasal resonance and nasal air emission, which had been recorded on patient charts. Resonance rating included normal; hypo-nasal; and mild, moderate or severe hyper-nasal. Intervention included surgical repair of the defect. The surgical repair was done by the researcher himself. The procedure performed included Von Langenback palatoplasty and V-Y pushback<sup>7</sup>. Post-Intervention assessment was done 6 weeks after surgery.

**Data Analysis:** Data was analyzed using SPSS Version-23 and statistical analysis performed. Descriptive statistics were used. Variables specially studied included age of palatoplasty, pre and

post-operative speech and VP gap. Chi-square test was applied to see any association of age group with speech results and velopharyngeal gap.  $P < 0.05$  was considered significant. Results were compared with literature and discussed.

#### RESULTS

Current study with a sample of 32 cases of isolated cleft palate with equal gender distribution revealed a mean age of  $1.4 \pm 0.63$  years at primary palatoplasty. Majority 26(81.3%) belonged to 1-4 years age group and 6(18.8%) were <1 years of age at the time of surgery. 18 (56.35%) cases had secondary cleft palate while 14 (43.8%) had primary cleft palate. Hyper-nasality and hyper-nasality with articulation errors were commonest symptoms in 12 (37.5%) cases each followed by regurgitation and Hyper-nasal voice with regurgitation in 4(12.5%) cases each. Langenback palatoplasty was most commonly performed procedure with 28 (87.5%) cases and good repair was achieved in 22 (68.8%) cases with 10 (31.3%) were left with large VP gap requiring pharyngoplasty (Table-I).

**Table-I: Operative & Post-Operative Clinical Features (N=32)**

Variable	Clinical Features	n (%)
Palatoplasty type	Langenback	28 (87.5%)
	V-Y pushback	4 (12.5%)
Palatoplasty result	Good	22 (68.8%)
	Velopharyngeal gap	8 (25%)
	Bifid uvula & Velopharyngeal gap	2 (6.2%)
Post-op speech & associated problems	Normal	14 (43.8%)
	Mild hyper-nasal	16 (50%)
	Hyper-nasal & articulation errors	2 (6.2%)
Post-op Vp gap*	Normal	20 (62.5%)
	Large	10 (31.3%)
	Small	2 (6.2%)

(Note: \*Vp gap = velopharyngeal gap)

Palatoplasty results as regards VP gap, when cross tabulated against cleft type (Table-II), revealed better repair results for secondary cleft with only 2 out of 18 cases post-operatively left with a large VP gap, while out of the 14 cases of primary cleft, 8 had large and 2 had small VP gap. This difference was statistically significant ( $p=0.002$ )

On the other hand when palatoplasty results were assessed for age association (Table-III), it revealed that there was no statistically significant association between speech results and age at palatoplasty ( $P=0.415$ ). As regards repair of plate in < 1 year age group, it revealed normal palatal repair in all, while in the 1-4 year age group out of 26 cases, 8 had post-operative VP gap while 2 had bifid uvula along with VP gap. However this difference was not statistically significant ( $P=0.187$ ). Also in the <1 year age group post-operative VP gap was normal while in the 1 to 4 year age group out of 26, 6 had large and 4 had moderate VP gap. This difference was statistically significant ( $P=0.012$ )

**Table-II: Outcome of Palatoplasty for Cleft type (N=32)**

Palatoplasty Outcome		n	Cleft type		X <sup>2</sup> , P-value
			Primary	Secondary	
Vp gap*	Normal	20	4	16	12.495 0.002
	Large	10	8	2	
	Small	2	2	0	
	Total	32	14	18	

(Note: \*Vp gap= velopharyngeal gap)

**Table-III: Outcome of Palatoplasty for Age Group (N=32)**

Palatoplasty Outcome		n	Age (years)		X <sup>2</sup> , P-value
			<1	01-04	
Speech and Associated symptom	Normal	14	4	10	1.758 0.415
	Hyper-nasal	16	2	14	
	Hyper-nasal voice & articulation errors	2	0	2	
	Total	32	6	26	
Repair	Good	22	6	16	3.357 0.187
	Vp gap	8	0	8	
	Bifid uvula & VP Gap	2	0	2	
	Total	32	6	26	
Vp gap*	Large	6	0	6	10.995 0.012 0.012
	Moderate	4	0	4	
	Normal	20	4	16	
	Small	2	2	0	
	Total	32	6	26	

(Note: \*Vp gap= velopharyngeal gap)

When pre-palatoplasty characteristics were cross tabulated against post palatoplasty speech (Table-IV), it revealed significant ((P=0.01) improvement in speech results compared to pre-operative speech. Also the improvement in speech following palatoplasty revealed significantly better results (P=0.007) for secondary cleft.

**Table-IV: Pre-Operative and Post-operative speech and associated symptoms (N=32)**

Pre-palatoplasty characteristics		Post-palatoplasty Speech			Chi-square association
Variable	Characteristics (n)	Normal	Hyper-nasal mild	Hyper-nasal & articulation errors	X <sup>2</sup> , p-value
Pre-op speech and associated symptoms	Regurgitation (4)	4	0	0	16.762, 0.01
	Regurgitation & hyper-nasal voice (4)	0	4	0	
	Hyper-nasality (12)	8	4	0	
	Hyper-nasality & articulation errors (12)	2	8	2	
	Total (32)	14	16	2	
Cleft type	Primary (14)	2	10	2	9.796, 0.007
	Secondary (18)	12	6	0	
	Total (32)	14	16	2	

**DISCUSSION**

Current study evaluated the results of palatoplasty in cases of isolated cleft palate, in terms of velopharyngeal gap closure and correction of speech with the hypothesis that delayed presentation results in VP gap/ insufficiency and poor speech

results. Delayed presentation being a norm in developing countries like Pakistan, hence a sample with mean age of 3.25 ± 2.55 years (81.2% being 1- 4 years old and 18.8% < 1 year old) with equal gender distribution facilitated the study with the following outcome:

To achieve optimum goal of cleft palate repair i.e. normal feeding and speech, closure of velopharyngeal gap is essential. Current section discusses the association of VP gap closure following palatoplasty with age at surgery. Present study revealed statistically significant association (P=0.012) between VP gap closure following palatoplasty with age at surgery. Though repair of plate in < 1 year age group, revealed normal VP gap in all, but in the 1-4 year age group out of 26 cases, 8 had post-operative VP gap while 2 had bifid uvula along with VP gap. In the same age group out of 26, 6 had large and 4 had moderate VP gap, while in the <1 year age group no case developed a large and moderate VP gap, supporting our hypothesis. In contrast in a study by Shaffer et al. velopharyngeal incompetence was not associated with age at palatoplasty<sup>8</sup>. Similarly, no age association was reported by Sullivan et al.<sup>9</sup> and following Furlow’s palatoplasty both before and after 5 years of age by Perkins et al.<sup>10</sup>. Other the other hand, Abdel-Aziz reported improved results with 85.7% with complete VP closure, in younger age group<sup>11</sup>. While in quite similarity to our study Annigeri et al. reported significant (<0.01) association of age with fistula formation with fistulae being less common in age group <2 years (8%) and very common above 2 years (66.6%)<sup>12</sup>, indicating that delayed surgical intervention does affect outcome and there is association between the VP gap closure and age at intervention especially in countries like Pakistan where delayed presentation is a norm.

Speech development is a more complicated process, which not only depends on the velopharyngeal closure but also on the functional status of the articulators and hence post-operative scarring of palatal soft tissue and impaired growth may affect the speech outcomes, hence timing of surgical intervention is an essential factor affecting speech outcome<sup>13</sup>. Hence we specially noted the association of speech outcome following palatoplasty with age at surgical intervention and results did not reveal any significant association (P=0.415) thus rejecting our hypothesis. However comparatively poorer results were noted in 1-4 years age group. Similarly Shaw et al., following a randomized trial comparing surgical intervention at 6 months and 1 year did not report any significantly improved speech outcome for earlier surgery<sup>14</sup>. In contrast Shaffer et al. reported that age at palatoplasty was significantly (<0.05) associated with speech language delay with those operated after 13 months of age were associated with more delay<sup>8</sup>. Similarly another author reported significant difference with only 16.6% having severe speech abnormalities for those operated before 2 years of age compared to 66.6% for those operated after 2 years of age<sup>12</sup>. Also good speech outcome was reported at younger age by Abdel-Aziz in a study using Furlow’s technique and reported normal nasality in 85.7% cases, mild hyper-nasality in 14.3% with speech improvement in those having mild hyper-nasality with no further surgical intervention<sup>11</sup>. Andreoli et al. also reported sufficient speech intelligibility following palatoplasty in >4 years

old patients<sup>15</sup>. However, in the present study post operatively, we found normal speech in 43.75%, mild hyper-nasality in 50% and hyper-nasality plus articulation errors in 6.25% cases and 31.3% required pharyngoplasty.

Although palatal repair does improve speech, its association with type of palatoplasty and timing of surgery is still not clear<sup>16</sup>, and requires further research. However, it must be emphasized here that as studies have used widely variable techniques to evaluate resonance and speech results, hence it is nearly impossible to compare results<sup>17</sup>.

**Limitations:** This study also has limitations due to the fact that only two types of surgical procedures were used for palatal closure and hence the results cannot be generalized. Bigger studies using all types of palatal repair procedures need to be conducted.

### CONCLUSION

Age at palatoplasty has significant association with velopharyngeal gap/ insufficiency ( $p=0.012$ ), while it has no significant association with speech results ( $p=0.415$ ) when comparing <1 year and 1-4 years age group.

### AUTHOR'S CONTRIBUTION

**Saqulain G:** Statistical analysis, Data interpretation, Manuscript final reading and approval

**Mumtaz N:** Conceived idea, Designed research methodology, Manuscript writing

**Ahmed J:** Data collection, Literature search

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### REFERENCES

- Burg ML, Chai Y, Yao CA, Magee W 3rd, Figueiredo JC. Epidemiology, Etiology, and Treatment of Isolated Cleft Palate. *Front Physiol.* 2016; 7:67. doi: 10.3389/fphys.2016.00067.
- Jajja MRN, Cawasji ZF, Imran S, Khan MS, Hashmi SS, Khan TS. Oral Clefts: A review of the cases and our experience at a single institution. *J Pak Med Assoc.* 2013; 63(9):1098-1102
- Ahmed J, Saqulain G. Frequency of otitis media with effusion in children presenting with cleft palate. *Rehab J.* 2017;01(02); 28-30.
- Thomas C. Repair of cleft palate: Evolution and current trends. *J Cleft Lip Palate Craniofac Anomal.* 2015;2:6-10.
- Téblick S, Ruymaekers M, de Castele EV, Nadjmi N. Effect of Cleft Palate Closure Technique on Speech and Middle Ear Outcome: A Systematic Review. *J Oral Maxillofac Surg.* 2019;77(2):405 doi.org/10.1016/j.joms.2018.09.027.
- Falzone SP. Optimal Age for Palatoplasty to Facilitate Normal Speech Development: What is the Evidence? In: Berkowitz S. (eds) *Cleft Lip and Palate.* Springer, Berlin, Heidelberg. 2006. Pp 691-703.
- Han HH, Kang IS, Rhie JW. Half-and-Half Palatoplasty. *Arch Craniofac Surg.* 2014;15(2):105-108. Doi: 10.7181/acfs.2014.15.2.105.
- Shaffer AD, Ford MD, Losee JE, Goldstein J, Costello BJ, Grunwaldt LJ et al. The Association between Age at Palatoplasty and Speech and Language Outcomes in Children With Cleft Palate: An Observational Chart Review Study. *Cleft Palate Craniofac J.* 2020;57(2):148-160. doi.org/10.1177/1055665619882566
- Sullivan SR, Vasudavan S, Marrinan EM, Mulliken JB. Submucous Cleft Palate and Velopharyngeal Insufficiency: Comparison of Speech Outcomes Using Three Operative Techniques by One Surgeon. *Cleft Palate Craniofac J.* 2011;48(5): 561-570
- Perkins JA, Lewis CW, Gruss JS, Eblen LE, Sie KC. Furlow palatoplasty for management of velopharyngeal insufficiency: a prospective study of 148 consecutive patients. *Plast Reconstr Surg.* 2005;116(1):72-80. doi: 10.1097/01prs.0000169694.29082.69. PMID: 15988249.
- Abdel-Aziz M. Speech outcome after early repair of cleft soft palate using Furlow technique. *Int J Pediatr Otorhinolaryngol.* 2013;77(1):85-88. doi: 10.1016/j.ijporl.2012.09.038.
- Annigeri VM, Mahajan JK, Nagarkar A, Singh SP. Outcome analysis of palatoplasty in various types of cleft palate. *J Indian Assoc Pediatr Surg.* 2012;17:157-161
- Peterson-Falzone S. Optimal Age for Palatoplasty to Facilitate Normal Speech Development: What is the Evidence?. In: Berkowitz S. (eds) *Cleft Lip and Palate.* Springer, Berlin, Heidelberg.2006; 691-703. https://doi.org/10.1007/3-540-30020-1\_45
- Shaw W, Semb G, Lohmander A. Timing of Primary Surgery for cleft palate (TOPS): protocol for a randomised trial of palate surgery at 6 months versus 12 months of age *BMJ Open* 2019;9:e029780. doi: 10.1136/bmjopen-2019-029780
- Andreoli ML, Yamashita RP, Trindade-Suedam IK, Fukushima AP. Speech intelligibility after primary palatoplasty: listener perception. *Audiol Commun Res.* 2016;21:e1650. Doi:10.1590/2317-6431-2015-1650.
- Schilling GR, Cardoso MDA, Maahs MAP. Effect of palatoplasty on speech, dental occlusion issues and upper dental arch in children and adolescents with cleft palate: an integrative literature review. *Revista CEFAC.* 201; 21(6); e12418. doi.org/10.1590/1982-0216/201921612418.
- Kummer AW, Hosseinabad HH, Redle E, Clark S. Protocols for Reporting Speech Outcomes following Palatoplasty or Velopharyngeal Surgery: A Literature Review. *Plast Reconstr Surg Glob Open.* 2019;7(2):e2151. doi: 10.1097/GOX.0000000000002151.