

Techniques and results of palate fistula repair following palatoplasty: a 234-case multicenter study

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ABSTRACT

Introduction. Palate fistula is the most frequent complication following palatoplasty. The objectives of this study were: to describe the most widely used repair techniques; to study results and recurrence rate; to analyze potentially predictive recurrence variables; and to assess whether a specific technique is superior according to fistula size and location.

Materials and methods. Retrospective study of patients undergoing palate fistula repair in 7 healthcare facilities from 2008 to 2018. All facilities had at least 20 new cases of cleft lip and palate annually (range: 20-80), with a fistula incidence of 14% (range: 1.5-20%). Minimum follow-up was 1 year. 8 variables were collected for statistical analysis purposes.

Results. 234 fistula patients underwent surgery. Most fistulas occurred in complete bilateral cleft lip and palate (Veau type IV). The most frequent location was the hard palate (Pittsburgh types IV and V (63.2%)), and fistulas were mostly large (42.1%) and medium (39.5%). The most frequent repair technique was re-palatoplasty (34.2%). Recurrence rate was 22%. The multivariate analysis demonstrated more recurrences in re-palatoplasty repaired type III fistulas in patients over 3 years old.

Conclusion. A tendency towards using flap repair in large hard palate fistulas, re-palatoplasty in medium hard palate and soft and hard palate junction fistulas, and local flaps or re-palatoplasty in small fistulas at any location was observed. However, it could not be statistically demonstrated whether a specific repair technique was superior in different clinical situations.

KEY WORDS: Cleft palate/surgery; Oral fistula/epidemiology; Oral fistula/surgery; Risk factors; Treatment results.

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This work was presented as a Medicine final degree project at San Pablo University Study Center (CEU).

Date of submission: March 2020

Date of acceptance: May 2020

TÉCNICAS Y RESULTADOS DE REPARACIÓN DE FÍSTULAS PALATINAS POSPALATOPLASTIA: ESTUDIO MULTICÉNTRICO DE 234 CASOS

RESUMEN

Introducción. La fístula palatina es la complicación más frecuente tras una palatoplastia. Los objetivos de este estudio fueron: describir las técnicas de reparación más frecuentemente empleadas; estudiar los resultados y la tasa de recidiva; analizar posibles variables predictivas de recidiva y valorar la posible superioridad de una determinada técnica según el tamaño y la localización de la fístula.

Material y método. Estudio retrospectivo de pacientes operados de fístulas palatinas desde 2008 hasta 2018 en 7 centros. Todos operaban al menos 20 casos nuevos de fisuras labiopalatinas al año (rango 20-80) con una incidencia de fístulas de 14% (rango: 1,5-20%). El seguimiento mínimo fue de 1 año. Se recogieron 8 variables para el análisis estadístico.

Resultados. Se operaron 234 pacientes con fístulas. La mayoría ocurrieron en fisuras labiopalatinas bilateral completa (tipo IV de Veau). La localización más frecuente fue el paladar duro (tipos IV y V de Pittsburgh (63,2%) y la mayoría fueron grandes (42,1%) y medianas (39,5%). La técnica de reparación más frecuente fue la repalatoplastia (34,2%). La tasa de recidiva fue del 22%. El análisis multivariante mostró más recidivas en fístulas tipo III reparadas con repalatoplastia, en mayores de 3 años.

Conclusión. Se observó una tendencia a utilizar más reparación con colgajo en fístulas grandes del paladar duro, repalatoplastia en fístulas medianas de paladar duro y de la unión, y colgajos locales o repalatoplastia en fístulas pequeñas en cualquier localización, pero no se pudo demostrar estadísticamente la superioridad de una técnica reparadora concreta en diferentes situaciones clínicas.

PALABRAS CLAVE: Fisura palatina/cirugía; Fístula oral/epidemiología; Fístula oral/cirugía; Factores de riesgo; Resultados de tratamiento.

INTRODUCTION

The main objective of cleft palate repair is to restore palate functional and physical integrity to allow for normal speech development and avoid regurgitation from the oral to the nasal cavity. Despite the correct use of various palatoplasty techniques, surgical closure disruption and subsequent fistula occurrence is not infrequent. Overall incidence, as published in a recent meta-analysis, is 8.6%⁽¹⁾, ranging from 4.7 to 60%⁽²⁾. Numerous palate fistula repair techniques following palatoplasty have been described⁽³⁾, but there are few result studies and currently there is no consensus regarding which is the most adequate according to fistula size and location⁽⁴⁾.

The objectives of this study were:

1. To describe the techniques most frequently used in palate fistula repair.
2. To study results and recurrence rate following repair.
3. To analyze potentially predictive fistula recurrence variables.
4. To assess whether a specific technique is superior according to fistula size and location.

MATERIALS AND METHODS

An observational retrospective study of all patients undergoing palate fistula repair following palatoplasty in 7 specialist healthcare facilities from 2008 to 2018 was carried out. All facilities had at least 20 new cases of cleft lip and palate annually (range: 20-80), with a fistula incidence following palatoplasty of 14% (range: 1.5-20%). Minimum follow-up was 1 year. Syndromic patients, persistent oronasal communication not repaired during primary palatoplasty, and bifid uvula were excluded. Table 1 features clinical record data.

Results were analyzed using the IBM SPSS Statistics software, version 22.0.0.0. <0.05 p values were considered

Table 1. Data collected from clinical records.

- Date of birth and sex
- Type of cleft palate, according to Veau classification⁽²³⁾:
I = soft palate, II = soft and hard palate, until the incisive foramen, III = complete unilateral cleft lip and palate, IV = complete bilateral cleft lip and palate
- Primary repair technique
- Age at palatoplasty
- Fistula location, according to Pittsburgh classification⁽²³⁾:
type I (bifid uvula), type II (soft palate), type III (soft and hard palate junction), type IV (hard palate), type V (primary and secondary palate junction in bilateral cleft lip and palate, Veau type IV), type VI (lingual alveolar) and type VII (labial alveolar)
- Fistula size, either large (>5 mm), medium (3-5 mm), or small (<3 mm)
- Technique used for fistula repair
- Occurrence of recurrence and persistence or non-persistence of symptoms

significant. The association among variables was studied using cross tables and Chi-square and Fisher's test.

RESULTS

A total of 234 patients with palate fistula following palatoplasty underwent surgery (132 boys (56%) and 102 girls (44%)). 30 children (13%) were under 3 years old at surgery, 87 (37%) were 3 to 6 years old, and 117 (50%) were over 6 years old. Primary cleft palate distribution according to Veau classification is featured in figure 1. More than half of them were complete bilateral cleft lip and palate (Veau type IV). The most frequent palatoplasty technique was the 2-flap technique, followed by the inverted double Z-plasty (Furlow's technique) (Table 2). Most cleft palates were repaired when the patient was 6-12 months old.

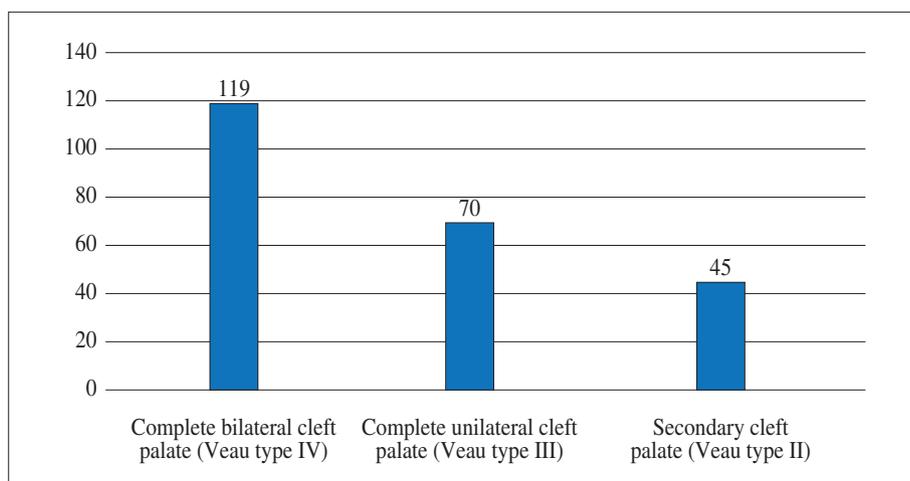


Figure 1. Primary cleft palate distribution in the cohort of patients undergoing palate fistula repair following palatoplasty.

Table 2. Primary palatoplasty techniques used in patients subsequently undergoing palate fistula surgery.

Technique	n	%
Lateral relaxation incision technique (Von Langenbeck)	30	13
Two-flap technique (Bardach and Wardill-Veau-Kilner variants)	119	51
Inverted double Z-plasty technique (Furlow's technique)	70	30
Others	15	6
Total	234	100

The most frequent fistula location was the hard palate (Pittsburgh types IV and V (148, 63%)), followed by the soft and hard palate junction (Pittsburgh type III (77, 33%)) and the soft palate (Pittsburgh type II (9, 4%)). 98 fistulas were large (42%), 92 were medium (39%), and 44 were small (19%). Most large (n = 60, 72%) and medium (n = 60, 67%) fistulas were located in the hard palate, and most small fistulas were located in the soft and hard palate junction (n = 24, 57%).

Table 3 features the techniques used for fistula repair. The most frequent technique was re-palatoplasty (82, 35%), followed by FAMM (Facial Artery Musculo Mucosal flap (61, 26%)), local flap with or without dermal matrix (60, 26%), and lingual flap (31, 13%).

The most widely used technique for large fistula repair was FAMM flap (n = 42, 43%); the most widely used technique for medium fistula repair was re-palatoplasty (n = 24, 26%); and the most widely used technique for small fistula repair was re-palatoplasty and local flaps (n = 15, 35%).

In terms of location, the most widely used technique for hard palate fistula repair was FAMM flap (n = 54, 37%); the most widely used technique for soft and hard palate junction fistula repair was re-palatoplasty (n = 33, 44%); and the most widely used technique for soft palate fistula repair was re-palatoplasty (n = 6, 66%).

Of the patients studied, 177 (76%) healed definitively, and 51 had surgical wound dehiscence and persistent fistula, with a recurrence rate of 22%. Half of those (n = 25, 11%) remained symptomatic.

The univariate analysis of predictive recurrence variables showed significant differences in terms of age at the primary palatoplasty, in terms of age at fistula repair, and in terms of fistula repair technique. Children aged 18-24 months old undergoing palatoplasty had a higher recurrence rate (40%, p <0.001) than those under 18 months old. Children under 3 years old undergoing fistula repair had a lower recurrence rate (10%, p <0.01), and re-palatoplasty demonstrated a significantly higher number of recurrences (33, 42%, p <0.001) than the other techniques.

Table 3. Techniques used in the 234 fistulas repaired.

Technique	n	%
Local flap	29	12
Local flap with dermal matrix	31	13
Re-palatoplasty	82	35
FAMM flap	61	26
Lingual flap	31	13
Total	234	100

Regarding fistula location, the multivariate analysis found a higher recurrence rate in re-palatoplasty repaired soft and hard palate junction fistula (Pittsburgh type III) (58%, p <0.001).

To sum up, even though flap repair tends to be more frequent in large hard palate fistulas, re-palatoplasty is typically more frequent in medium hard palate and soft and hard palate junction fistulas, and local flap repair and re-palatoplasty are usually more frequent in small fistulas at any location. No specific repair technique statistically proved to be superior in the different clinical situations.

DISCUSSION

Palate fistula is the most frequent complication following cleft palate surgery⁽⁵⁾. Potential causes include insufficient tissue mobilization, closure under tension, inadequate postoperative pain management, and hematoma⁽⁶⁾. Once in place, the fistula can be symptomatic or asymptomatic, causing persistent regurgitation, hygiene difficulty with frequent halitosis, and hypernasal speech. Asymptomatic fistula does not require treatment, but symptomatic fistula represents an additional surgical challenge, since clinical signs are very diverse, previous scars make re-interventions difficult, there is little local tissue, and there is no consensus regarding the best repair technique. Fistula incidence rates in the literature vary widely^(7,8). Studying fistula rate following palatoplasty was not the objective of this study. Healthcare facilities performing more than 20 primary cleft palate surgeries annually were included, using the same nomenclature and classification systems, and with a fistula incidence below 20%.

Similarly to other studies^(9,10), no sex differences were found in the number of fistulas or recurrence rate. Other authors have associated the risk of developing fistulas with primary cleft palate severity⁽¹¹⁾, which is consistent with our findings. Most fistulas were secondary to Veau type IV cleft palate repair, followed by type III and type II cleft palate. Lithovius et al.⁽¹²⁾ found no association with the primary cleft palate repair technique used. However,

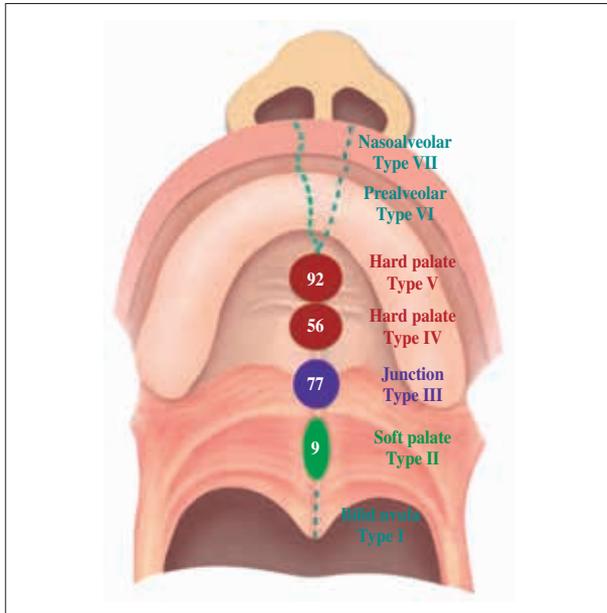


Figure 2. Palate fistula location.

Cohen et al.⁽⁷⁾ and Garg et al.⁽¹³⁾ found a higher fistula incidence following 2-flap techniques and in patients over 2 years old undergoing surgery. In our series, the largest number of fistulas was also found with the 2-flap technique

(Table 2). This can be due to the superimposed suture lines used in this technique, whereas in Furlow's technique, nasal sutures are covered by oral sutures and vice-versa. In addition, Furlow's technique is usually carried out in narrower cleft palates. Primary cleft palate width was not studied in this work.

The most frequent fistula location was the hard palate (Pittsburgh IV and V), followed by the soft and hard palate junction (Pittsburgh III), consistent with other studies^(13,14) (Figs. 2-4). The fact the nasal plane is more difficult to dissect and the oral fibro-mucosa is less distended is probably accountable for these differences. This could also explain that fistulas are larger and more frequent in this location.

To repair the 234 fistulas of this series, a wide range of techniques described in the literature were used⁽²⁻⁶⁾ (Table 3). Given that most fistulas were large (>5 mm) and medium (3-5 mm), the most frequent techniques involved either a wide detachment/cleavage and palate tissue mobilization (re-palatoplasty), or the provision of regional vascularized tissue (FAMM flap). Re-palatoplasty provides a nice fistula visualization, facilitates wide tissue mobilization, allows for a new closure with less tension, and can be combined with lengthening techniques⁽¹⁵⁾. However, fistula recurrence rate remains relatively high, since less vascularized palate scar tissue is used.



Figure 3. 8-year-old patient undergoing complete bilateral cleft lip and palate (Veau type IV) surgery. The fistula was asymptomatic, it caused liquid and solid regurgitation, and it contributed to hypernasality and halitosis. Pre- and post-surgery appearance of a large (>5 mm) Pittsburgh type V hard palate fistula, undergoing surgery with left, anterior pedicle, reverse flow Facial Artery Musculo Mucosal (FAMM) flap.



Figure 4. 14-year-old patient undergoing complete unilateral left cleft palate (Veau type III) surgery. Pre- and post-surgery appearance of a large (>5 mm) Pittsburgh types IV and V hard palate fistula, undergoing 2-flap re-palatoplasty with retroposition of the velar muscles and lengthening for velopharyngeal insufficiency in a single surgical maneuver.

Therefore, for large hard palate fistulas, particularly with significant scar fibrosis, many authors suggest using regional flaps providing vascularized tissue, such as FAMM flap⁽¹⁶⁻¹⁸⁾ or lingual flap⁽¹⁹⁾. Both have demonstrated good results, but in a comparative study, Sohal et al. concluded that FAMM flap involves fewer early complications, such as traumatic dehiscence and hematoma, less difficulty to eat and speak, and shorter operating times⁽²⁰⁾. Kocaaslan et al. reported traumatic separation in 9 out of 34 patients undergoing lingual flap surgery, and recommended it is not carried out in patients under 7 years old, since post-operative cooperation tends to be poorer. For large palate fistulas occupying virtually the whole hard palate, various authors suggest using thin extraoral free flaps – radial fascia lata flap⁽²¹⁾ or transverse ulnar forearm flap (TUFF)⁽²²⁾. This indication is more frequent in post oncologic surgery defects in adults. However, in cleft palate patients, it can occur following complete necrosis of the palatoplasty flaps. In our 234-case series, none of the fistulas required free flap closure. For small fistulas, local flaps can be used, but they are typically difficult to mobilize, and complete sealing is hard to achieve. This is why some authors have suggested combining them with dermal matrix, as it was the case in 31 cases in our series, with good results⁽²³⁾.

The techniques used allowed for definitive closure in nearly 80% of cases, with a 22% recurrence rate, similar to that described in the literature^(2,6,23).

Predictive fistula recurrence variables were similar to those described in the literature for fistula occurrence: being over 18 months old at palatoplasty, and being over 3 years old at fistula repair and 2-flap re-palatoplasty, particularly in soft and hard palate junction fistulas (Pittsburgh III). These same factors are probably accountable for such differences, since older children have firmer tissues, with larger scars and less vascularized borders, especially in the area of maximum tension in the soft and hard palate junction. Regarding palatoplasty, not only are fistula occurrence and recurrence lower when repaired in patients under 18 months old, but also functional integrity is provided at the right time of speech development. However, lower refistulization risks when fistula repair is performed in patients under 3 years old should be weighed against the possibility of performing another surgical maneuver in the treatment protocol, especially if the fistula is small and little symptomatic.

CONCLUSION

A tendency towards using regional flaps in large fistulas, especially in the anterior hard palate (Pittsburgh V), re-palatoplasty in medium fistulas in the hard palate and in the soft and hard palate junction, and local flaps with or without dermal matrix in small fistulas at any location was observed in the cohort studied. According

to the multivariate analysis, no technique proved superior to others in terms of fistula size or location. Children under 3 years old undergoing fistula surgery had lower recurrence rates.

REFERENCES

1. Hardwicke JT, Landini G, Richard BM. Fistula incidence after primary cleft palate repair: a systematic review of the literature. *Plast Reconstr Surg.* 2014; 134: 618e-27e.
2. Sullivan SR, Marrinan EM, LaBrie RA, Rogers GF, Mulliken JB. Palatoplasty outcomes in nonsyndromic patients with cleft palate: a 29-year assessment of one surgeon's experience. *J Craniofac Surg.* 2009; 20 Suppl 1: 612-6.
3. Denny AD, Amm CA. Surgical technique for the correction of postpalatoplasty fistulae of the hard palate. *Plast Reconstr Surg.* 2005; 115: 383-7.
4. Denadai R, Zanco GL, Raposo-Amaral CA, Buzzo CL, Raposo-Amaral CE. Outcomes of surgical management of palatal fistulae in patients with repaired cleft palate. *J Craniofac Surg.* 2020; 31: e45-e50.
5. Saralaya S, Desai AK, Kumar N. Difficulty index-based management of palatal fistula after primary cleft palate repair: An institutional experience. *J Oral Maxillofac Surg.* 2019; 77: 851. e1-e7.
6. Bonanthaya K, Shetty P, Sharma A, Ahlawat J, Passi D, Singh M. Treatment modalities for surgical management of anterior palatal fistula: Comparison of various techniques, their outcomes, and the factors governing treatment plan: A retrospective study. *Natl J Maxillofac Surg.* 2016; 7: 148-52.
7. Cohen SR, Kalinowski J, LaRossa D, Randall P. Cleft palate fistulas: A multivariate statistical analysis of prevalence, etiology, and surgical management. *Plast Reconstr Surg.* 1991; 87: 1041-7.
8. Emory RE, Jr, Clay RP, Bite U, Jackson IT. Fistula formation and repair after palatal closure: An institutional perspective. *Plast Reconstr Surg.* 1997; 99: 1535-8.
9. Garg R, Shah S, Uppal S, Mittal RK. A statistical analysis of incidence, etiology, and management of palatal fistula. *Natl J Maxillofac Surg.* 2019; 10: 43-6.
10. Rautio J, Andersen M, Bolund S, Hukki J, Vindenes H, Davenport P, et al. Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate: 2. Surgical results. *J Plast Surg Hand Surg.* 2017; 51: 14-20.
11. Landheer JA, Breugem CC, van der Molen AB. Fistula incidence and predictors of fistula occurrence after cleft palate repair: Two-stage closure versus one-stage closure. *Cleft Palate Craniofac J.* 2010; 47: 623-30.
12. Lithovius RH, Ylikontiola LP, Sándor GK. Incidence of palatal fistula formation after primary palatoplasty in Northern Finland. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 2014; 118: 632-6.
13. Lu Y, Shi B, Zheng Q, Hu Q, Wang Z. Incidence of palatal fistula after palatoplasty with levator veli palatini repositioning according to Sommerlad. *Br J Oral Maxillofac Surg.* 2010; 48: 637-40.
14. Murthy J. Descriptive study of management of palatal fistula in one hundred and ninety-four cleft individuals. *Indian J Plast Surg.* 2011; 44: 41-6.

15. Elsherbiny A, Grant JH 3rd. Total palatal mobilization and multi-lamellar suturing technique improves outcome for palatal fistula repair. *Ann Plast Surg.* 2017; 79: 566-70.
16. Rossell-Perry P, Arrascue HM. The nasal artery musculomucosal cutaneous flap in difficult palatal fistula closure. *Craniofac Trauma Reconstr.* 2012; 5: 175-84.
17. Bénateau H, Garmi R, Chatellier A, Ambroise B, Maltezeanu A, Veyssièrre A. Palato velar fistulae in cleft palate. *Ann Chir Plast Esthet.* 2019; 64: 406-12.
18. López-Sánchez R, Berenguer-Fröhner B, González-Meli B, Rodríguez-Urcelay P, Marín-Molina C, de Tomás-Palacios E, Núñez-Villaveirán T. Colgajo FAMMM para reconstrucción de fistulas de paladar en pacientes con fisurapalatina congénita: experiencia y resultados. *Cir Plast Iberolatinoam.* 2014; 40: 261-70.
19. Gupta N, Shetty S, Degala S. Tongue flap: a “workhorse flap” in repair of recurrent palatal fistulae. *Oral Maxillofac Surg.* 2020; 24: 93-101.
20. Sohail M, Bashir MM, Khan FA, Ashraf N. Comparison of clinical outcome of facial artery myomucosal flap and tongue flap for closure of large anterior palatal fistulas. *J Craniofac Surg.* 2016; 27: 1465-8.
21. Cavadas PC, Pérez-Espadero A, Rubí Oña CG, Thione A. Prelaminated fascia lata free flap for oronasal fistula reconstruction. *Plast Reconstr Surg Glob Open.* 2017; 5: e1262.
22. Martin MC, Machado GR, Wong WW, Yeung LC, Jean RD. Transverse ulnar forearm flap. *J Craniofac Surg.* 2010; 21: 1741-4.
23. Agir H, Eren GG, Yasar EK. Acellular dermal matrix use in cleft palate and palatal fistula repair: A potential benefit? *J Craniofac Surg.* 2015; 26: 1517-22.