Original Article

FREQUENCY OF FISTULA FORMATION AFTER TWO STAGE REPAIR OF CLEFT PALATE

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ABSTRACT

Objectives: *To estimate the frequency of fistula formation after two stage repair of cleft palate.*

Materials and Method: *A total of 50 patients having completed cleft palate repair with or without lip repair were recruited from December 2007 to February 2009. Patients were diagnosed on history and oral examination. Informed consent was taken from parents. Width of the cleft measured preoperatively. Two stage repairs were done by a single surgeon with a time lapse of six months.*

Results: *Five patients developed clinically significant oronasal fistula. All these fistulas occurred at the junction of hard palate and soft palate within 3 weeks time.*

Conclusion: *The overall rate of true fistula development was 12% over a mean follow up period of 6 months. The incidence of true fistulas that were symptomatic and subsequently required surgical repair was 10%. Fistula rates were higher for more wide clefts but were not affected by gender or age.*

Key words: *Palatal Fistula, Oronasal Fistula, Cleft Palate, Cleft Lip and Palate.*

INTRODUCTION

Palatal cleft is a relatively common facial disorder, characterized by separation of the palatal segments and a resultant open communication between the mouth and nose.[1] Cleft palate is one of the commonest congenital abnormalities with a worldwide incidence of 1 in 700.[2] It either occurs as an isolated cleft palate, which is genetically and morphologically separate entity or as a cleft lip or palate (CL/P).[1,3] CL/P is frequent in Asian while infrequent in African American.[2,3] Cleft palate alone has an incidence of 0.45-0.5/1000 births. The prevalence of other anomalies in cleft lip patients is estimated to be 7-13% where as patients with CL/P have been reported to have 11-14% chance of having other anomalies at birth.[4] In addition to abnormal facial appearance, these patients have got significant difficulties in their social interactions, their ability to communicate effectively and difficulties in eating and hearing.[5]

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In 1958 Kernahan et al[6] based their classification on embryology, dividing palate into primary and secondary. According to this classification primary palate is that segment of the palate which is anterior to the incisive foramen while secondary palate is the part which is posterior to incisive foramen. A cleft of the secondary palate is further divided into complete or incomplete depending on the involvement of soft or hard palate.[3,6,7] Smiths modification of Kernahan classification is in common use because it is simple to understand and more users friendly.[3,7,8]

The goals for repairing a cleft palate are restoration of normal speech, hearing and normal facial growth for the infant. In managing cleft palate, the most controversial issues are the timing of surgery, speech development and its effect on facial growth.[3,9,10,11] The ideal age for cleft palatal surgery is usually 3 to 18 months. Speech and hearing are improved by cleft palate repair before twenty four months of age. Delayed closure (after five years) is associated with retardation of maxillofacial growth.[3,12,13]

The most significant complications after palatal repair are wound dehiscence and oronasal fistula. A cleft palate fistula is any palatal defect posterior to incisive foramen resulting from failure of healing or a

breakdown in the primary surgical repair of the palate.[10,14,15] Oronasal fistula has an incidence of 5-29%.[16] Fistula formation after repair depends on surgeon's experience, width of cleft, type of cleft, technique, wound tension, single layer repair, dead space below the mucoperiosteal flap and maxillary arch expansion. According to size, fistulae have been classified as small (1-2 mm), medium (3-5 mm) and large (> 5 mm). Schultz has divided these fistulae into pinpoint, slit, oval and total dehiscence.[15]

We descriptively evaluated our post palatoplasty patients with the aim to find the frequency of cleft palatal fistula after two stage repair of cleft palate as no such local study is available covering major issues or aspect on which to base problem solving. As severity of cleft greatly affects the development of postoperative palatal fistula, so the parents of the patients with wide cleft can be briefed before surgery regarding the prognosis following the repair.

MATERIALS AND METHOD

This observational descriptive study was conducted at the department of Plastic Surgery Postgraduate Medical Institute, Hayatabad Medical Complex, Peshawar from December 2007 to February 2009. A total of 50 cases were included using non probability purposive sampling technique.

Inclusion criteria:

- All the patients with complete cleft lip and palate and isolated cleft secondary palate as well.
- Patients undergoing two staged palate repair as it is the standard procedure followed in our unit.
- Patients who have undergone Furlow's repair for soft palate in our unit and came for Langenbeck's repair for hard palate as second stage.

Exclusion criteria:

- Patients with partial or sub mucus cleft palate.
- Patients undergoing operation other than the above mentioned repair such as single stage repair.
- Fistulae or unrepaired clefts in the alveolar region.
- Patients who underwent Furlows repair in other units.

All the cases were diagnosed on the basis of history and clinical examination of oral cavity. Necessary investigations were performed. The parents of the patients were briefed about the congenital anomaly, treatment modalities and timing of surgery. Informed consent was taken from parents. Size of the cleft was measured per operatively and all patients underwent two stage repair. Both procedures were done under general anesthesia using an endotracheal tube, supplemented with local infiltration of lignocaine 0.5% with 1:100,000 epinephrine. Furlow's repair was done for cleft soft palate at the age of 3-9 months and Von Langenbeck's repair for cleft hard palate at the age of 12-18 months as this is the standard procedure performed in our unit.

Patients were given plenty of fluids postoperatively to keep the mouth moist and clean. All patients were given prophylactic antibiotics postoperatively along with oral analgesics in the form of suspension. Fluid diet was given using spoons or straws. Patients were discharged on the third day when patients have started taking orally adequately. Patients were then followed up at two weeks, three and six months respectively. Both procedures were done by a single surgeon. The data was collected on a Performa, and was analyzed using SPSS version 10.

RESULTS

Fifty patients were included in this study. Sixty-two percent of the patients were male while 38% were female with male to female ratio of 1.63:1. Age range was 6 to 19 months with a mean age of 13.64$\pm$3.14 months in both sexes respectively (Table 1). Three cases had a positive family history of cleft lip and palate. The minimum width of cleft was 0.8 cm and maximum 1.8 cm, with a mean value of 1.25 $\pm$ 0.3177 cm. Unilateral cleft lip and palate was observed in 24 cases (48%) while bilateral cleft lip and palate was observed in 13 cases (26%). Isolated cleft palate was present in 13 cases (26%) as shown in Table 2. Cleft lip and palate may occur in association with other congenital anomalies and was seen in 03 cases. The number of patients who developed clinically significant oronasal fistula was 5 (Table 3). Fistula sizes ranged from 2-4 mm. Fistula location was at the junction of soft and hard palate which occurred within 3 weeks. Five patients with fistula required surgical repair, thus reflecting a true frequency of 10%. Fistula rates were higher for patients with wide clefts but were not affected by

gender. The secondary surgery was higher in patients with more severe clefts. The severity of cleft was determined by measuring the width of cleft palate. Sixteen cases (32%) were wide (> 1.5 cm) and 34 cases (68%) were narrow (<1.5 cm). Other complications seen were bleeding 6% and wound dehiscence 8% as shown in Table 4. Frequency of complications was higher in those patients having bilateral CL/P.

Table 1: Age and sex distribution for CL/P

	n	Mean Age in months	SD
Male	31	13.48	3.33
Female	19	13.89	2.88
Total	50	13.64	3.14

Table 2: Types of Cleft Palate

	n	%
Unilateral cleft palate	24	48
Complete cleft palate	13	26
Bilateral cleft palate	13	26
Total	50	100

DISCUSSION

The primary goal in the timing of cleft palate repair is to provide adequate palatal function for the normal speech development without any detrimental effect on maxillofacial growth.[3] Early repair of palate increases the likelyhood of normal speech development.[3] Delayed treatments may interfere less with maxillofacial growth but speech development to be effected adversely. Previously surgeons advocated delayed closure of the entire palate or early repair of the soft palate only and management of hard palate prosthetically for several years. The patient treated with this protocol had almost normal facial growth but had significant irreversible speech problems.[9,17] Clefts that are 1.5 cm or more in width at the junction of soft and hard palate have a statistically significant risk of fistula formation.[5,7] The most difficult area for closure is around the junction of the hard and soft palate which is the most frequent site for the formation of a palatal fistula postoperatively.

Table 4: Complications of two stage palatoplasty

Complications	n	%
Oronasal fistula	5	10
Wound dehiscence	4	08
Bleeding	3	06

Facial and palatal deformation as a consequence of repair is due to destruction of blood supply and scar formation.[17] To avoid these consequences, some surgeons advocate two stage approach to palate repair, with early repair of the soft palate only and later repair of the hard palate.[14] This protocol entailed repair of the soft palate, around 4 to 6 months, at the same time as lip repair. The hard palate was obturated and later repaired at about 4 to 5 years of age. An earlier age has been proposed subsequently for hard palate repair.[7]

The ideal age for palate repair is 3-6 months as suggested by Kaplan,[3] based on the theory that the palate must be functional when the sounds related to the palate are first learned just to avoid any detrimental effects on speech development and integration. Hence an early two stage palate repair is being advocated in the management of patients with cleft lip and palate. The sequence involves the Furlows repair for cleft lip and soft palate at 3 to 6 months of age with secondary closure of hard palate at 15 to 18 months of age by Von Langenbecks technique. The postop-

Table 3: Characteristics of patients with Fistula

No.	Age in Months	Type of Cleft	Width (cm)	Fistula size (mm)	Time of occurrence
1	12	Bil. CL/P	1.8	3x4	3rd Week
2	12.5	Unil. CL/P	1.5	2x3	3rd Week
3	14	Compl. CL/P	1.8	2x2	3rd Week
4	16	Unil. CL/P	1.8	2x2	3rd Week
5	13	Bil. CL/P	1.8	2x2	3rd Week

erative restricted mobility of soft palate for another 3 to 6 months has been attributable to postoperative edema which settles spontaneously and thus palate can function normally till 9 to 12 months of age.

In this study there were 6 fistulae, out of which 5 were clinically significant and definitely needed repair. All of these patients were of male gender. These fistulas occurred in clefts where the width was more than 1.5 cm. This finding suggests that chance of fistula formation is higher as width of cleft increases. These findings have been supported by other international studies such as those of Muzaferr[14] who concluded that low rate of clinically significant fistula (8.7%) is attributed to early soft palate repair, with smaller secondary clefts allowing repair with minimal dissection and disruption of vascularity. Mak et al[18] and Phua et al[19] also reported that fistula rate were higher in patients having wider clefts and were not affected by the type of surgical repair.

Our results were comparable to other international studies[14,15,18] held in various centers and stays with the same opinion that 2 stage repair yields an excellent speech results with an acceptable fistula rate. Furlows in stage 1 addresses two important factors, i.e. reconstruction of functional sphincter and velar stretch/length, mandatory for an excellent soft palate repair.[3] A straight mid line scar is also avoided with this Z-plasty technique which prevents postoperative shortening in the anteroposterior direction. Theoretically, it permits reorientation of the soft palate musculature from an anteroposterior direction to a transverse direction. This subsequently facilitates more normal muscular function. The Furlows procedure is primarily a technique of soft palate closure. The hard palate repair can be performed by a variety of techniques preferably including Von Langenback or Veau-Wardill-Killner push-back technique. Khozla[20] attributed excellent speech result with an excellent fistula rate of 2.17% to Furlows double opposing Z- plasty technique. The fistula rates were not related to operating surgeon, gender, age, technique and use of presurgical orthopedics or palatal expansion. Another factor responsible for fistula formation in our study was delayed closure of the cleft, i.e. after 12 months of age as most of cleft palate centers advocate cleft repair before the patient is one year of age, except in unusual circumstances such as Pierre Robin sequence, Treacher Collins, Apert, Crouzons or other craniofacial syndromes in which patients are at risk of airway obstruction and oronasal fistula formation. In these situations it is wise to delay repair until the child is 2 years of age.[21]

Emory et al[22] reported that the strongest predictor of the occurrences of oronasal fistula was the surgeon performing the procedure and extent of cleft has no effects. In our study, a single surgeon carried out the operation so the outcome of our study can be linked to the experience of operating surgeon. Despite an important role of presurgical orthopedics and orthodontics, the patients in this study could not avail this facility because of its high cost. The results were reflected in the clinical outcome of our palatoplasties. Recently, Schendel et al[23] reported very low incidence of palatal fistula formation (3.2%) using Delaire palatoplasty while Wilhelmi et al[24] reported a rate of 3.4% using two flap palatoplasty. Inspired by these reports Mak et al[18] became interested by Furlows palatoplasty. Fistula rate came out to be 5.6% within the first three years. Furlows palatoplasty was adopted as standard procedure.[18,23] Similar results have been shown by Chen and Noordhoff,[25] who reported 4 oronasal fistulas in 35 patients undergoing Furlows Palatoplasty. This reflects the importance of surgeons experience in accomplishing favorable clinical outcome.[19]

In spite of various techniques available for repair of cleft palate and management of fistula, fistula formation will continue to occur even in hands of best surgeons and in best centers. A multidisciplinary team approach is the best way in managing these patients with cleft palate and fistula.

CONCLUSION

The management of a patient with cleft palate and fistula is multidisciplinary. Such a team should include a Plastic Surgeon, an Otorhinolaryngologist, an Oral Surgeon, an Orthodontist, a Dentist, a Speech therapist, an Audiologist, a Geneticist, a Nurse coordinator, a Social worker and a Psychologist. For this purpose a center for these patients should be established where all these facilities should be available under one roof. The incidence of true fistulas that were symptomatic and subsequently required surgical repair was 10%. Fistula rates were higher for more wide clefts but were not affected by gender or age.

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