

## Frequency of palatal fistula following 2 stage palate repair

Muhammad Shadman, Adeeba Ahmad, Aneela Ambreen, Tahmeedullah, SM Haider, Zarar Niazi, Uzma, Waqas Hayat, Imran, Nasir

### Abstract

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

**Material and Methods:** This observational descriptive study was carried out at department of plastic and reconstructive surgery PGMI, Hayatabad Medical Complex, Peshawar from December 2013 to February 2015. A total of 50 patients having complete cleft palate with or without lip were selected. Patients were diagnosed on the basis of history and oral examination. Informed consent was taken from parents. Width of the cleft was measured preoperatively. Two steps repair was done by a single surgeon with a time lapse of six months.

**Results:** 6 patients developed clinically significant oro-nasal 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 developments 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.

**Keywords:** Palatal fistula, oro-nasal fistula, cleft palate, cleft lip and palate.

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**Mardan Medical Complex, Peshawar**  
M Shadman  
Z Niazi  
**Khyber Teaching Hospital, Peshawar**  
SM Haider  
**Hayatabad Medical Complex, Peshawar**  
A Ahmad  
Tahmeedullah  
Uzma  
Imran  
Nasir  
**Naseer Teaching Hospital, Peshawar**  
A Ambreen

### Correspondence:

Dr Muhammad Shadman, SR, Plastic and Reconstructive Surgery, Mardan Medical Complex, Peshawar.  
Cell: + 92-300-5876918  
Email: shadmanch@gmail.com

### Introduction:

Cleft palate is one of the commonest congenital abnormalities with a worldwide incidence of 1 in 700.<sup>1</sup> It either occurs as an isolated cleft palate, which is generally and morphologically separate entity or as a cleft lip or palate (CL/P). (CL/P) is frequent in Asian while infrequent in African American. Isolated cleft palate shows a constant ratio of 0.45-0.5/1,000 births. About 7-13% of patients with isolated cleft lip and 11-14% of patients with CL/P have other anomalies at birth.<sup>2,3,4</sup>

It has tremendous aesthetic and functional implications for patients in their social interactions, particularly on their ability to communicate effectively and on their facial appearance in addition to their eating and hearing difficulties.<sup>3</sup>

Numerous classifications have been suggested over the years because of diverse complexity

of the cleft lip and palate. The first generally accepted classification was developed by Davis and Ritchie.<sup>5,6</sup>

Kernahan subsequently proposed a visual classification in 1971, the striped Y classification which is the most widely adopted classification in use.<sup>7</sup> We preferably use Smiths modification of Kernahan classification because it is simple to understand and more users friendly. The goals of a surgeon repairing a cleft palate are normal speech, normal facial growth and hearing for the affected infant.<sup>4,8</sup>

The complications of great concern in the post-operative period in addition to bleeding and respiratory distress are wound dehiscence and oro-nasal 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.<sup>9</sup>

The incidence of clinically significant oro-nasal fistula being 04-34% or even more in cases of primary palate repair.<sup>10,11,12</sup> While others concluded the rate of fistula as 05-29%.<sup>13</sup> An oro-nasal fistula of secondary palate may occur following palatoplasty even in the hands of expert surgeons.<sup>12</sup> The management of a fistula secondary to cleft palate repair is limited in success, with a high incidence of recurrence after initial fistula closure. Factors that may contribute to fistula formation are type of cleft, type of repair, wound tension, single layer repair, dead space below the mucoperiosteal flap and maxillary arch expansion. In general, the more severe the cleft, the more likely it is that a post-operative palatal fistula will occur. Fistulae have been classified according to size, small 1-2 mm, medium 3-5mm and large greater than 5mm.<sup>9,10,14</sup> Pittsburgh classification is based on the site of fistula as type 1: bifid uvula; type 2: soft palate; type 3: Junction of the soft and hard palate; type 4: Hard palate; type 5: Junction of primary and secondary palate; type 6: Lingual alveolar and type 7: Labial Alveolar.<sup>15</sup>

As a number of patients come for cleft palate repair to our unit, our aim was to study the frequency of cleft palatal fistula in these patients as no such local study is available covering major issues or aspect on which to base problem solving. Also we will in a position to get the guidelines regarding patient selection and the technique as well.

#### **Objective:**

The objective of my study was to estimate the frequency of fistula after two stage repair of cleft palate.

#### **Material and Methods:**

After approval of study by institutional ethical committee a total of 50 patients with complete cleft lip and palate, isolated cleft secondary palate, and patients who have undergone Furlow's repair for soft palate in our unit and for same Langenbeck's repair for hard palate as second stage were selected. Patients with partial or sub-mucous cleft palate, patients undergoing operation other than the above mentioned procedure, fistulae or unclosed clefts in the alveolar region

and patients who underwent Furlows repair in other units were excluded from the study.

The patients were diagnosed on the basis of history and examination of the oral cavity. Informed consent was taken from parents. Size of the cleft was measured pre-operatively. Prophylactic antibiotics were given to all patients. Both procedures were done under general anaesthesia 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. Furlow's repair of soft cleft palate was done using double opposing Z-plasties from the oral and nasal surfaces. While in Von Langenbeck's repair, bipedicle mucoperiosteal flaps were created by incising along the oral side of the cleft edges and along the posterior alveolar ridge from the maxillary tuberosities to the anterior level of the cleft. The flaps were then mobilized medially with preservation of the greater palatine arteries and closed in layers.

Patients were given plenty of fluids postoperatively to keep mouth moist and clean. All patients were given antibiotics post-operatively 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.

Since the time lapse between two stages was six months with total followed up time of six months so the study was completed in one year. Both procedures were done by a single surgeon.

The data was collected on a proforma, and was analysed using SPSS version 10.

#### **Results:**

Out of 50 patients only 6 developed clinically significant oro-nasal fistula. Fistula sizes ranged from 1mm to 5mm. All these fistulas were lo-

Table-1: Characteristics of patients with fistula

No	Age in months	Type of Cleft	Width (cm)	Fistula (mm)	Time of Occurrence
1	12	Bil. CL/P	1.8	3x4	3rd week
2	12.5	Uni. CL/P	1.5	2x3	3rd week
3	14	Compl. CL/P	1.8	2x2	3rd week
4	16	Uni. CL/P	1.8	2x2	3rd week
5	13	Bil. CL/P	1.8	2x2	3rd week

Table-2: Frequency of fistula

Fistula	Frequency	Percent
Yes	05	10.0
No pie chart	45	90.0
Total	50	100.0

Table-3: Age and sex distribution for CL/P

	n	Mean	Std.deviation
Age	50	13.64	3.14
Male	31	13.48	3.33
Female	19	13.89	2.88

Table-4: Frequency of fistula

	Frequency	Precent
Unilateral cleft palate	24	48
Complete cleft palate	13	26
Bilateral cleft palate	13	26
Total	50	100

Table-5: Associated anomalies with CL/P

	Frequency	Percent
Positive	03	06
Negative	47	94
Total	50	100

cated at the junction of soft and hard palate and occurred within 3 weeks (table-1). Number of patients with fistula requiring surgical repair was 05 reflecting a true frequency of oro-nasal fistula as 10% (table 2). 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.

Majority of the patients were male. Maximum age of patients at the time of repair was six months and maximum age of 19 months. (table 3) Family history of cleft lip and palate was present in 03 cases. Minimum width of cleft was 0.8 and maximum 1.8 cm, with a mean value of 1.25cm and SD of 0.3177. (table-1)

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 in table 4.

Patients were observed for any early and late complications referring to figure-1 and 2. Early complications seen were bleeding 03 cases and wound dehiscence 04 cases. Frequency of early and late complications was higher in those patients having wider clefts (>1.5cm) figure-3.

**Discussion:**

The primary objective of cleft palate repair is to restore the anatomy of palate as close to normal as possible, provide an adequate mechanism for speech and improve middle ear function.<sup>16</sup> The most effective technique for cleft palate repair continues to be debatable.<sup>17</sup> In general, all techniques use some form of mucoperiosteal flap for the hard palate closure, reconstruction of muscular velopharyngeal sling in soft palate and approximation of tissue without tension to accomplish near to normal functional outcome.<sup>16</sup>

A lot of variables have an effect on outcome of surgical repair of cleft palate. Cohen suggested in his study that fistula formation was surgeon dependent and to a lesser extent depends on technique and type of cleft.<sup>18</sup>

Very wide clefts are difficult to close. Clefts that are 1.5cm or more at the junction of soft and hard palate have statistically significant risk of fistula formation. The goal of obtaining complete nasal and oral closure from front to back may not be possible in wide clefts. The most difficult area for closure is around the junction of the hard and soft palate which is the frequent site for the formation of a palate fistula postoperatively.

Facial and palatal deformation as a consequence of repair is due to destruction of blood supply and scar formation.<sup>16</sup> Some surgeons intended to overcome these consequences, advocate two stage approach to palate repair, with early repair of the soft palate only and later repair of the hard palate. This protocol originally introduced by Schweckendiek, entailed repair of the soft pal-

ate around 4 to 6 months at the same time as lip repair. The hard palate was obdurate and later repaired at about 4-5 years of age. A bit earlier age has been proposed subsequently for hard palate repair.<sup>5</sup>

The rationale for his approach has been that the hard palate cleft narrows during the interval between two stages, requiring less dissection resulting in less maxillary disturbance and eventually leading to good functional outcomes. Hence an early two stage palate repair is being advocated in the management of patients with cleft palate and lip. The sequence involves the Furlows repair for cleft of soft palate at 3-6 months of age with secondary closure of hard palate at 15-18 months of age by Von Langenbecks technique.

There were 6 fistulae in our study out of which 5 were clinically significant and definitely needed repair. All of these patients were of male gender. A total of 5 fistulae occurred in clefts having widths greater than 1.5 cm suggesting that chance of fistula formation is higher as width of cleft increases. The concept of wide cleft isn't vague or irrelevant but has bearing on postoperative oronasal fistula formation. These findings have been supported by other international studies such as those of Muzaferri et al 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.<sup>9</sup>

Another study done by Anthony Sze-Yuen Mak et al, suggested that postoperative fistula formation is related to the size of initial defect. Yun Shan Phua and Tristan de Chalain also concluded that fistula rate were higher in patients having wider clefts and were not affected by the type of surgical repair.<sup>19,20</sup> Our results were comparable to other international studies held in various centers and stays with the same opinion that two stage repair yields an excellent speech results with an acceptable fistula rate. Furlows in stage one addresses two important factors i.e. reconstruction of functional sphincter and velar length, mandatory for an excellent soft palate repair.<sup>4</sup> A straight mid line scar is also avoided with this Z-plasty technique which prevents postoperative shortening in the antero-posterior direc-

tion.

Charles L et al attributed excellent speech result with an excellent fistula rate of 2.17% to Furlows double opposing Z-plasty technique.<sup>21</sup> Another factor for fistula formation in our study was delayed closure of the cleft i.e. after 12 months of age in unusual circumstances as Pierre Robin sequence, Treacher Collins, Apert, Crouzons or other craniofacial syndromes in which patients are at risk of airway obstruction and oro-nasal fistula formation.<sup>22</sup>

Stephen Bresnick et al carried out a study regarding the correlation between Treacher Collins syndrome and post-operative fistula formation. He attributes it to an abnormal vascular supply to the palate in patients with Treacher Collins syndromes.<sup>23</sup>

Children undergoing Furlows repair at children hospitals Los Angeles from 92 children with other two syndromes and cleft palate and 458 nonsyndromic children with isolated cleft palate were studied children with Treacher Collins syndrome had significantly higher fistula rates (50%) than children with other syndrome (8.7%) or no syndrome (4.1%).<sup>23</sup> Despite an important role of pre-surgical orthopaedics and orthodontics our patients in our series couldn't avail this facility because of its high cost. The results were reflected in the clinical outcome of our palatoplasties.

Recently very low incidence of palatal fistula formation from a single surgeon experience has been reported (3.2%) by Schended using Delaire palatoplasty (3.4%) and by Wilhelmi using two flap palatoplasty.<sup>24,25</sup> Similar results have been shown by Chen and Noordhoff, who reported 4 oronasal fistulas in 35 patients undergoing Furlows palatoplasty. This reflects the importance of surgeon experience in accomplishing clinical outcome.<sup>26</sup> Nevertheless the commonest site of oro-nasal fistula formation is junction of hard and soft palate.<sup>27</sup>

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. The best way of management of the patient with cleft

palate and fistula is multi-disciplinary team approach.

#### Conclusion:

We conclude that the overall rate of 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. The management of a patient with cleft palate and fistula is multi-disciplinary. Such a team should include a plastic surgeon, an Otorhinolaryngologist, an oral surgeon, an orthodontist, a dentist, a therapist, an audiologist, geneticist, a nurse coordinator, a social worker and a psychologist. Although good results can be accomplished without a team approach, but better results can be assured should be established where all these facilities should be available under one roof.

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#### Role and contribution of authors:

Dr Muhammad Shadman concept and study design, conducted research, data collection interpretation and analysis, drafting and final approval of paper

Dr Adeeba Ahmad manuscript writing, data collection and its analysis and interpretation of results

Dr Aneela ambreen drafting, data collection and analysis, literature review, critical review of manuscript for important intellectual content

Dr Tahmeedullah, concept and study design, data analysis and interpretation of results, critically reviewed the manuscript for intellectual content

Dr Syed Muhammad Haider collected the data and references and helped in introduction writing

Dr Zarar Niazi, collected the data and references and helped in the discussion writing

Dr Uzma, collected the data and references and helped in interpretation of data

Dr Waqas Hayat, collected the data and references and helped in discussion writing

Dr Imran, collected the data and references and helped in result writing

Dr Nasir, collected the data references and helped in result and conclusion writing.

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