

# Palatal Fistula Post-Cleft Palate Repair: A Tertiary Center Experience in Oman

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## Abstract

One of the known complications of cleft palate surgery is development of fistula. This study highlights our experience with cleft palate surgery in relation to fistula occurrence at our center. This is one of the first studies of this kind in Oman. We retrospectively reviewed 362 records of cleft palate patients. The most common technique used for cleft palate repair at our center was Veau-Wardill-Kilner technique. In our series we have noticed palatal fistulae in 32% of cases. This high rate is partially due to inclusion of very anterior fistulas in patients with complete cleft lips and palates, which actually represent part of the alveolar cleft rather than breakdown of the palatal repair. This will be closed during time of alveolar bone grafting at a later stage. We recommend a future prospective controlled study to study the factors that lower the incidence of fistula in our population.

## Keywords

Palate, Palatoplasty, Palatal Fistula, Post-Operative Complication

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## 1. Introduction

The management of Cleft Palate needs involvement of a multidisciplinary team which includes Plastic surgeons, otolaryngologists, orthodontists and speech language therapists. Surgical repair of cleft palate is important for feeding, normal speech mechanism and hearing. The recommended age for palate repair is between 6 - 12 months [1] and repair at 18 months of age is practiced by some [2]. The surgical goals for palate repair are complete palatal closure, velopharyngeal competence, and near normal maxillary growth. There are various factors that may contribute in the prevention of palatal fistula includes adequate mobilization and tension free closure especially in wide clefts (wider the cleft, higher the fistula rate), good three layer closure irrespective of the technique

used, proper realignment of the muscle of the soft palate in the mid line, and postoperative management.

The development of a fistula is one of the well-known complications that might develop after a surgical repair [3]. The incidence of fistula reported in a systematic review in cleft lip and palate cases was higher (17.9%) than cases of cleft palate alone (5.4%) with an over-all incidence of 8.6%. The development of fistula might occur due to a number of factors such as age at operation, type and extent of clefting in addition to the surgical technique used [3] [4]. This research aims at studying the various factors that are associated with development of cleft palate. Gender difference appears to be one of the reported factors in literature. Males had a greater incidence of cleft lip and/or palate [5]-[11]. In contrast, females had more severe cases [5] [7]. On the other hand, a study done in Netherlands found no significant association between fistula development and age, gender or cleft type [12]. The effect of the surgical repair on maxillary growth is still controversial. However there are some factors that affect the maxillary growth for example the interventions done before surgery, cleft repair timing, technique of the surgery and post-operative protocol [13]. It has been reported that Veau-Wardill-Kliner technique of palatal repair shows a good mid-facial result in the long term [14]. The study aims at deciding the best surgical technique that is associated with the lowest rate of fistula. Furlow palatoplasty and two-flap palatoplasty are some of the surgical techniques described in the literature to be associated with low fistula rate (5.5%) [15].

## 2. Methods: Ease of Use

### Selecting a Template

#### Methods

Patients with cleft lip and/or palate operated at Khoula Hospital between the period of January, 2008 and July, 2015 were included in the study. Patients with cleft lip only were excluded from the study.

Veau classification was used to classify cleft palate cases (**Table 1**).

Fistula cases were diagnosed upon follow-up clinically on examination or based on history of nasal regurgitation when drinking. Our fistula cases included very anterior fistulas which are close to alveolar clefts since we depend on the patients' record files. There was no distinction and no clear differentiation made in the exam note between alveolar clefts and fistula.

Patient's data was extracted from the hospital "Al Shifa 2" system (Khoula

**Table 1.** Veau classification.

Veau classification	Description
Veau I	Defects of the soft palate only
Veau II	Defects involving the hard and soft palate
Veau III	Defects involving the soft palate to the alveolus usually involving the lip
Veau IV	Complete Bilateral Clefts

Hospital). The data was analyzed using the “Statistical Package for Social Sciences” (SPSS) version 19 for Windows.

Cases were reviewed twice independently by the first two authors to extract fistula cases.

### 3. Results

Total of 362 cases were included in the study 182 (50.3%) male and 180 (49.7%) female patients. Mean age of those patients at time of surgery was 28.27 months (range 2.0 - 432.0 months old; Standard Deviation (SD) 53.74 months). **Table 2** shows the age distribution according to gender.

Distribution of cleft palate cases was as follows: 88 cases (24.3%) were Veau I, 156 (43.1%) of cases were Veau II, 83 (22.9%) cases Veau III and 35 (9.7%) of cases were Veau IV (**Figure 1**).

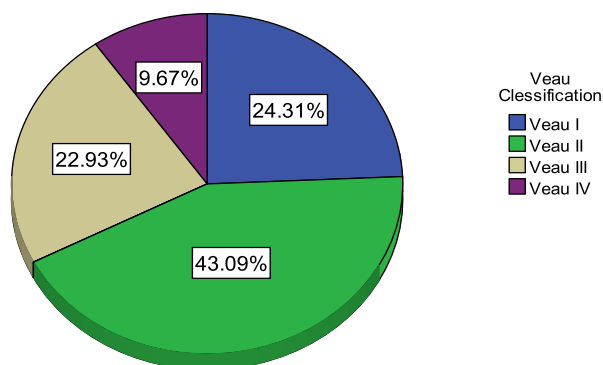
Veau-Wardill-Kliner technique was used to repair 270 (74.6%) of the cases. In 84 (23.2%) cases, Von Langenback technique was used. While Bardach Salyer technique was used in only 5 (1.4%) cases and Furlow repair in 3 (0.8%) cleft palate cases. **Table 3** shows the different techniques used for different types of clefts.

Palatal fistula has developed in 118 (32.6%) of operated cases noting that 40 (11%) of cases has failed to follow up. Mean time frame between surgery and fistula appearance was 8.77 months with a range of 1 to 78 months post-operation and SD of 13.51 months. **Table 4** shows the distribution of fistula cases according to the type of surgical repair.

Chi square was used to investigate any relation between surgical technique used and appearance of fistula. It showed a p-value of 0.77%, which meant that there was no significant association between surgical technique used and appearance of fistula.

Of the cases that developed fistula, 59.3% (70 out of 118 cases) were males. The increase in male cases was statistically significant with a p-value of 0.041%. Odds ratio for male to female risk of developing a fistula was 1.641 (**Table 5**).

**Extent of clefting using Veau Classification**



**Figure 1.** Extent of Clefting using Veau Classification of patients included in this study.

**Table 2.** Mean ages and Cleft type distribution according to Gender.

	Gender		Total	
	Male	Female		
Mean Age at time of surgical repair (months)	26.962	29.583	28.265	
Standard deviation of the age at time of surgery	54.8440	52.7121	53.7360	
Extent of clefting (Veau Classification)	Defects of the soft palate only	35	53	88
	Defects involving the hard and soft palate	81	75	156
	Defects involving the soft palate to the alveolus usually involving the lip	49	34	83
	Complete Bilateral Clefts	17	18	35
	Total	182	180	362

**Table 3.** Surgical technique used in different types of cleft palate.

	Surgical technique				Total
	Veau-Wardill-Kilner Technique	Von Langenback Technique	Bardach Salyer Technique	Furlow repair (Double oppsing Z-plasties)	
Veau I: Defects of the soft palate only	61	24	1	2	88
Veau II: Defects involving the hard and soft palate	95	60	0	1	156
Veau III: Defects involving the soft palate to the alveolus usually involving the lip	80	0	3	0	83
Veau IV: Complete Bilateral Clefts	34	0	1	0	35
Total	270	84	5	3	362

**Table 4.** Fistula Development distribution in relation to sex.

		Gender		Total	
		Male	Female		
Fistula Development (Yes, No, Lost follow up)	Yes	Number of cases	70	48	118
		% within Fistula cases	59.3%	40.7%	100.0%
		% within Gender	38.5%	26.7%	32.6%
	No	Number of cases	96	108	204
		% within Fistula cases	47.1%	52.9%	100.0%
		% within Gender	52.7%	60.0%	56.4%
	Lost follow up	Number of cases	16	24	40
		% within Fistula cases	40.0%	60.0%	100.0%
		% within Gender	8.8%	13.3%	11.0%
	Total	Number of cases	182	180	362
		% within Fistula cases	50.3%	49.7%	100.0%
		% within Gender	100.0%	100.0%	100.0%

**Table 5.** Distribution of fistula according to type of surgical repair.

	Fistula Development			Total
	Yes	No	Lost follow up	
Veau-Wardill-Kilner Technique	88	142	28	258
	34.1%	55.0%	10.9%	100.0%
Von Langenback Technique	28	57	11	96
	29.2%	59.4%	11.5%	100.0%
Bardach Salyer Technique	1	3	1	5
	20.0%	60.0%	20.0%	100.0%
Furlow repair (Double opposing Z-plasties)	1	2	0	3
	33.3%	66.7%	.0%	100.0%
Total	118	204	40	362
	32.6%	56.4%	11.0%	100.0%

Correction of palatal fistula was planned in 43 (35.6%) of fistula cases. Repair using local flaps was done in 24 (20.3%) of total fistula cases. Veau-Wardill-Kliner technique was used to repair 4 (3.4%) fistula cases. Only one case (0.8%) repaired primarily. While 2 fistula cases (1.7%) has failed to follow up, no repair was done in 87 (73.7%) of fistula cases within the study period and were advised to continue follow up (Table 6). Table 7 shows the surgical techniques used to repair the fistulae.

#### 4. Discussion

The main goals of the surgical repair are to restore the normal anatomic alignment evidenced by no nasal regurgitation during food intake and to achieve a good functional outcome evidenced by good speech production.

Many centers have reported their cleft palate surgery outcomes in the literature. The cases of distribution between males and females was equal [3]. Other studies showed more cleft palate cases among males [5]-[11]. Higher incidence of isolated cleft palate cases was observed in females [11] [16]. Higher Prevalence of cleft lip and palate cases was noted than cases of cleft palate or lip only [17] [18].

Our study demonstrated a mean age of patients at time of operation of 28.27 months of age with a wide SD. Our current practice at Khoula hospital is age of approximately 12 months at time of operation. The obtained result can be due to late presentation of some cases.

Distribution of cases favored Veau II, followed by Veau I, Veau III and Veau IV.

Our study demonstrated that there was no significant association between surgical technique used and appearance of fistula. The distribution of cases on different surgical repair types might have an effect on the result. Veau-Wardill-Kliner technique is the most commonly used technique at our institution. This might be a possible limitation of our study that is attributed to the nature of the study as it is a retrospective study.

**Table 6.** Surgical repair of cases of fistula

	Fistula developed			Total
	Yes	No fistula	Lost follow up	
Yes, operation planned and/or done	42	1	0	43
No, operation not planned	74	203	2	279
Lost follow up	2	0	38	40
Total	118	204	40	362

**Table 7.** Surgical technique used in cases of fistula

	Fistula Developed			Total
	Yes	No fistula	Lost follow up	
Local Flap	24	0	0	24
	20.3%	0.0%	0.0%	6.6%
No repair done	87	204	4	295
	73.7%	100.0%	10.0%	81.5%
Lost Follow up	2	0	36	38
	1.7%	0.0%	90.0%	10.5%
Primary repair	1	0	0	1
	0.8%	0.0%	0.0%	0.3%
Veau-Wardill-Kliner Technique	4	0	0	4
	3.4%	0.0%	0.0%	1.1%
Total	118	204	40	362
	100.0%	100.0%	100.0%	100.0%

In comparison with a study that compared association of fistula with rotation palatoplasty and conventional palatoplasty a statistically significant difference between the two groups. The conventional palatoplasty group included Furlow palatoplasty and Veau Wardill Kliner techniques. Overall fistula appearance was 17.7%, in which 6% (4/67) was in the rotation palatoplasty group and 18% (18/100) was in the conventional palatoplasty group [19].

A study done in university of Texas Southwestern Medical center compared patients with and patients without palatal fistula. They reported no significant difference between the two groups in regard to gender, age at palatoplasty and type of palate repair [20].

Palatal fistula rate excluding international unrepaired anterior palatal fistula at the alveolus in a retrospective study of cases repaired by one surgeon using Von Langenback Technique was 8 patients out of 177. Surgical repair was required in 2 cases out of those fistulae.

Higher incidence of fistulae was demonstrated in cases of bilateral cleft lip and palate [3]. A study done in New Zealand reported fistula rate of 12.8% (27 out of 211 children) [3]. Another study done in Canada at University of Toronto and University of Alberta showed a fistula rate of 0% (out of 148 and 34 operated

cases respectively). They used Modified Von Langenbeck technique in their study [21]. A study compared one-flap with two-flap palatoplasty showed no statistical significance in terms of fistula development [22]. A study which focused on studying delayed cleft palate repair after 18 months in adopted patients showed 33 % incidence of velopharyngeal insufficiency compared with 13% in the group repaired between 6 to 12 months of age [1].

Our study demonstrated a high rate of fistula development. There was no significant association between surgical technique used and development of fistula. However, there was significant increase in cases of fistula in males.

A meta-analysis done by Bykowski and others showed a significant association between Veau classification and fistula development. Fistula cases were more among Veau IV cases [23]. Another study showed the same association between fistula development and Veau class IV in a retrospective study of cases done by one surgeon [24].

At Khoula hospital we follow a strict post-op cleft palate feeding protocol. The protocol states liquid diet feeding for 5 days followed by 2 weeks of soft diet.

A contributing factor to the failure of cases to follow up is the fact that Khoula hospital is a tertiary plastic center that deals with cases from all across the country. Problems of transport and the distance between area of residence and our hospital might limit the ability of patients to continue to follow up. Another factor is that appointments are around 6 months to a year apart. Some parents might simply forget about the appointment. Some parents might be satisfied with their results and will stop following up.

Knowing the factors associated with palatal fistula post-operatively will help us reduce its incidence or even prevent it. Hence reduce the number of operations need to repair the fistula, decrease follow-up appointments and improve overall patient's satisfaction.

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## 5. Limitations

Our study faced a number of limitations beginning with the fact that the palate repair was done by multiple surgeons with different techniques and different level of experience. Since it is a retrospective study, we depended on the documentation and records of patient's files. The extraction of data was difficult in some cases as the exams findings and detection of fistulae differs with different surgeons especially if the fistula is very small and does not pose any clinical symptoms. Our study had an overestimated number of fistulae due to the fact that we included anterior palatal fistulae in continuity with alveolar clefts. The reason for that is that our exam notes did not clearly distinct between alveolar clefts and fistulae. Some of the most anterior fistulae which are in continuity

with alveolar cleft can be corrected at the time of alveolar bone grafting. We did not study the palatal width as it was not measured and could not be traced from the records although it was found as non contributing factor to fistula appearance in some studies [24]. The site of fistula formation was not included in our study as it was not mentioned in most of the patient's records.

## 6. Conclusion

Closure of fistula located at any part of human body is a surgical challenge so as Palatal fistula. No such study was conducted in Oman to show the incidence of Palatal fistula after primary palate repair. The aim of this study was not only to know the incidence but also the contributing factors which leads to this complication which is very difficult to treat. By knowing and evaluating the factors associated with development of palatal fistula, help us to standardize the surgical techniques to prevent/reduce the incidence of fistula formation. This helps to improve the overall results by avoiding the need for another anaesthesia, hospital stay , number of operations need to repair the fistula, decrease follow-up appointments and improve overall patient's satisfaction. By keeping this retrospective study as a baseline we can proceed with prospective studies after making the appropriate strategies and protocols for the repair of cleft palate to obtain better results.

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