Experience of unilateral cleft lip repair using the Anatomical Subunit Technique in an Omani population over a 5 year period

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Abstract

Objectives: The Anatomical Subunit Technique for unilateral cleft lip repair has gained popularity worldwide. The purpose of this study is to report our experience using this technique. Methods: 114 consecutive cases of cleft lip underwent primary cleft lip repair with closed rhinoplasty by a single surgeon from 2015 to 2020. In addition to the demographic data, severity and type of the cleft lip, the surgical outcomes, including vermillion notching, were assessed by an independent senior surgeon and rate of revision surgery were collected from Al-shifa plus 3 health electronic system. Parents’ satisfaction regarding scar quality, lip and nose aesthetic appearance was collected anonymously. The documented data were evaluated using statistical analysis. Results: 82 cases satisfied the inclusion criteria. The mean age at surgery for cleft lip was 32 weeks. 35 cases (43%) were complete cleft lips and 47 cases (57%) were incomplete. 43 children (52%) were born of consanguineous marriage. Six patients (7%) needed revision surgery. The digital survey was completed by 40 subjects and showed 85% satisfaction rate with the postoperative scar, 77.5% satisfaction with the aesthetic appearance of nose. Conclusion: In our hands, the anatomical subunit technique resulted in a predictable result with high patient’s satisfaction rate as related to scar quality, nasal and lip symmetry in children with varying severity of cleft lip. Revision for vermilion
excess was needed in 7% of cases. The high percentage of consanguinity (52%) in our study highlight the need for more targeted national campaigns involving premarital counselling in the Omani population.

**Keywords:** Cleft Lips, cheiloplasty, Oman, Surgical Technique, Millard Technique, Fisher, Consanguinity.

**Advances in Knowledge**
- This is the first study in Oman to highlight the outcome and direct experience of this technique for cleft lip repair in an Omani population.
- Exploring the direct opinion and experience of parents whom children has underwent cleft lip repair using the above technique.
- Presenting demographic data in a population of Omani patients with cleft lip.

**Application to Patient Care**
- The Anatomic Subunit Technique in our hands, produced predictable and identical results as related to scar quality, nasal and lip symmetry in children with varying severity of the cleft lip.
- Our study revealed high percentage of consanguinity (52%) in the study group, calling for more targeted national campaigns involving premarital counselling in the Omani population.
- The most common complication associated with anatomical subunit technique in our study was vermilion excess, which was corrected with simple wedge excision.

**Introduction**
Orofacial clefts remain the most common craniofacial congenital anomaly.¹,² A study published in 2001 by Rajab and Thomas concluded that the prevalence of cleft lip in Oman was 15 per 10,000 births.³ Out of those 0.62 per live births were combined cleft lip and palate, and 0.34 per live births were isolated cleft lip cases.³ This result is striking as it suggests a higher prevalence of cleft lip in Oman in comparison to the worldwide numbers. The Data published from the *International Perinatal Database of Typical Oral Clefts* in 2011 stated that the worldwide prevalence of cleft lip was found to be 9.9 per 10,000 births,⁴ in comparison to 15 per 10,000 births in the Sultanate.³ The incidence of cleft lip has been found to be associated with consanguineous marriages, specifically in children born to parents who are first cousins with a percentage of 61.5%, this incidence falls down to 18.0%
in parents who are second cousins.\textsuperscript{5} A meta-analysis study of orofacial clefts has indicated a clear relationship between consanguinity and non-syndromic orofacial clefts (NSOFC), with the risk being twice in children born to consanguineous parents.\textsuperscript{5}

Cleft lip anomalies are more commonly found in association with cleft palate deformities, however, in other less common instances, it can present as an isolated cleft lip anomaly.\textsuperscript{1,8} Cleft lip anomalies can be unilateral or bilateral and can range in severity from a scale of a microform cleft lip which is considered the least disfiguring to complete cleft lip.\textsuperscript{1,6} Some of the well-known challenges of children born with cleft lip include feeding difficulties, trouble in articulation and speech development, along with psychological and self-esteem concerns related to the condition.\textsuperscript{1} Studies related to the comprehensive statistics of this anomaly and its management outcomes in Oman remain significantly limited.

This is a retrospective study for the surgical correction of the cleft lip anomaly in an Omani population using a single technique by a single surgeon over a period of 5 years. Our institute, Khoula hospital is a level-1 trauma centre and is also the main plastic surgery centre in the Sultanate of Oman. The standard technique used for correction of cleft lip in our institute has been Millard or modified Millard Rotation Advancement Technique. The Anatomic Subunit Technique was introduced to the department of plastic surgery by the senior author in 2015.

Different studies have reported their experience with anatomical subunit technique in comparison to Millard and Modified Millard technique. Deshmukh et al,\textsuperscript{7} published a study favouring the aesthetic outcomes of Fisher’s technique over Mohler’s technique.\textsuperscript{7} In the study, 50 patients were selected and randomly assigned to undergo surgical cleft lip repair by either the Mohler or Fisher technique performed by a single surgeon.\textsuperscript{7} Outcomes were then assessed by laymen and demonstrated that a bigger proportion of evaluators have favored the aesthetic outcomes of the Fisher repair.\textsuperscript{7}

Kwang et al\textsuperscript{10} also published a similar study, which enrolled candidates from different levels of training and requested them to evaluate the aesthetic outcomes of anatomical subunit repair in comparison to rotation advancement repair.\textsuperscript{10} In this study, candidates from different levels of training have found outcomes of the anatomical subunit techniques to be
aesthetically superior to that of the latter technique.\textsuperscript{10} The experience of the anatomic subunit technique has not been studied in the Omani population prior to this study.

**Methods**

The Ethical approval was obtained from the Ministry of Health, Centre of Studies and Research at the Sultanate of Oman, Research Ethics Committee, Khoula Hospital. Verbal consent was taken from the parents whom were interested in filling the electronic survey and for those willing to include their children’s photos in this paper.

Inclusion criteria: All children who had underwent cleft lip repair from July 2015 to April 2020 using the Anatomic Subunit Technique of cleft lip repair at Khoula hospital, Muscat, Oman. Exclusion criteria: Children who lost follow up, parents who refused to be part of the study due to social or cultural reasons, children with a microform cleft lip, children with bilateral cleft lip, inadequate data entered in the Al-shifa plus 3 health electronic system.

All the patients included in the study were operated by a single surgeon from 2015 to 2020 in department of plastic surgery at Khoula hospital, Muscat, Oman, over a period of 5 years for varying degree of cleft lip ranging from unilateral incomplete cleft lip to complete cleft lip, using the anatomic subunit technique of cleft lip repair.

All the patients were evaluated preoperatively in the clinic and prioritized according to the age and anesthetic clearance for surgery. Naso-alveolar molding by experienced orthodontist was requested for indicated cases. Most of the children were operated by the 5\textsuperscript{th} to 7\textsuperscript{th} month of age after adequate weight gain, and hemoglobin evaluation. Once the children were cleared by the pre-anesthetic clinic for surgery, they were admitted and operated on the following day. All the children underwent cheiloplasty using the anatomic subunit technique with closed rhinoplasty by a single surgeon under general anesthesia. The procedure starts with markings as per the anatomical subunit technique then the key landmarks were tattooed. Infiltration of xylocaine 1 % with 1:200,000 adrenaline in the alar base and inferior turbinate area on the cleft side are performed. Incisions are made on the medial lip along the markings including the opening triangle. A cuff of the orbicularis oris muscle is dissected free after releasing its attachments to the Columella base and alveolar cleft margin. Next, the lateral lip incisions are made along the markings and after discarding the tissue along the cleft margin, a cuff of the orbicularis oris muscle was dissected free in the same manner. Next, the alar base
on the cleft side is released off the pyriform and advanced anteromedially. Closure started with the nasal floor and in some cases a turbinate flap is used to cover any exposed periosteal surface of the pyriform margin. Primary closed rhinoplasty is done with repositioning of the cleft side dome using 5-0 Monocryl. The mucosa, the muscle and the lateral vermillion flap are each approximated with 5-0 Vicryl, and the skin is closed with 7-0 Prolene. Intra-operative nostril retainer was applied in all cases and secured using a 4/0 nylon to the membranous septum. Elbow splints were applied and maintained for 10-14 days. The children were allowed breast feeding 3-4 hours post operatively and discharged on the 1st or 2nd postoperative day. The children were re-evaluated at 3 weeks, 6 weeks, 3 months, 6 months and then annually in the clinic. The average long-term follow-up for patients in our study was 2.3 years. The postoperative evaluation of all children who underwent cleft lip repair included evaluation and assessment of the quality of the scar on the lip and vermillion, symmetry of nose, symmetry of the vermillion volume and white roll alignment by an independent senior surgeon.

Parents of all the children who had undergone cleft lip repair using the anatomic subunit technique of cleft lip repair at Khoula hospital were sent a link of electronic survey regarding the aesthetic appearance of the lip after surgery, ability to breast feed, parents’ satisfaction with surgical scar and with correction of the nasal deformity. Data was collected from Al-Shifa plus 3 health electronic system of department of plastic surgery at Khoula hospital, Muscat, Oman. The data included those of personal data, date of surgery, severity and side of cleft lip, presence of a protruding premaxilla, pre-surgical orthodontics, grade of cleft palate, associated syndromes, the geographical area child from, associated anomalies, consanguinity, family history of cleft, last follow up date, objective assessment and any revision surgery for the lip. The data were entered in excel sheets and analyzed using IBM SPSS statistics program version 24.

Results
A total of 114 cleft lip cases were performed by the senior author during the study period. Out of 114 patients, 82 have satisfied the inclusion criteria and were included in our study. Out of the included cases 52 were males (63%), and 30 were females (37%). The mean age at the time of surgery was 32 weeks. Nine cases (11%) have undergone the surgery at later weeks due to the late presentation and co-morbid conditions. The eldest of our patients has presented for the first time at 356 weeks due to personal and socioeconomic issues, and was
operated at the age of presentation. A total of 43% of the cleft cases were from Muscat and Ad Dakhiliya governorates with 18 (22%) and 17 (21%) cases, respectively. The other regions made up 57% of the cases with Ash Sharqiah and Al Batinah representing 20% and 19% respectively. Ad Dhahirah, Al Wusta, Mustandam and finally Al Buraimi accounting for 11%, 4%, 2% and 1% respectively. All cleft lip cases were repaired by anatomical subunit technique.

Thirty-Five cases (43%) were found to be complete cleft lip and 47 cases (57%) were found to be incomplete. 53 cases (65%) were left sided and 29 cases (34%) were right sided involvement. 36 patients (44%) had cleft of palate involving both hard and soft palate, out of which 28 patients (34%) had a Veau’s class 3 cleft palate, 8 patients (10%) had Vaeu’s class 2. Twenty-one patients (26%) underwent pre-surgical naso-alveolar molding and 3 patients had protruding premaxilla. Twelve patients had associated anomalies, which were mainly cardiac anomalies. Two children were confirmed as syndromic, one was Downs syndrome and the other was Velo-Cardio-Facial syndrome. Forty-three children (52%) were born of consanguineous marriage [Figure 5], 33 were first cousin marriage (77%), 7 were second cousin marriage (16%), 3 were third cousin marriage (7%). 22 patients (27%) had a positive family history of cleft. Six patients (7%) out of the 82 who had undergone cleft lip repair by the anatomic subunit technique needed revision [Figure 6] as judged by the independent senior surgeon at the time of last follow up during the study period, 5 of which were for the vermilion excess, that was managed with simple wedge resection. A digital survey was sent to all the 82 parents, whose children had undergone cleft lip repair by anatomical subunit technique out of which 40 parents had responded to the survey anonymously. Analysis of the data obtained showed 82.5% (33/40) satisfaction with the aesthetic appearance of lip following cleft lip repair by anatomical subunit technique, 85% (34/40) were satisfied with the postoperative scar over the lip, 77.5% (31/40) were satisfied with the aesthetic appearance of nose following cleft lip surgery with closed rhinoplasty.

**Discussion**

Various techniques have been used by plastic surgeons to correct cleft lip anomalies,9,11 all of which aimed to establish the best functional rehabilitation and aesthetical acceptance.11-13 Rose’s straight-line repair was one of the first described techniques to repair cleft lip, this technique was published in 1891, followed by the rotation advancement technique for unilateral cleft lip repair which was introduced by Dr. Ralph Millard in 1955, this technique
is initiated by forming an incision just below the nostril which continues to run vertically to the philtral ridge.\textsuperscript{13-15} Millard’s technique has undergone a number of refinements throughout the years but probably remains the most common technique used in unilateral cleft lip repair worldwide.\textsuperscript{16,17} Mohler further modified the rotation-advancement repair technique in 1987, achieving a more symmetric positioning of the scar. The next big evolution in cleft lip repair came with the introduction of extended Mohler cleft lip repair by Court Cutting in 2003.\textsuperscript{19} The major adjustment of this technique was to extend the incision to the noncleft philtral column.\textsuperscript{12} With this modification, two major shortcomings have arisen, first was the complex appearance of the scar under the nasal sill which did not blend with the anatomical landmarks, second was that in order to avoid the under rotation and elevation of the cupid’s bow peak, the lateral lip segment was shortened.\textsuperscript{12}

To address the mentioned shortcomings, Dr. David M. Fisher proposed the anatomic subunit technique first published in 2005.\textsuperscript{12} As the name suggests, this technique respects the anatomical subunits of the lip and is based upon accurate pre-operative measurements.\textsuperscript{12} This technique preserved the transverse length of the lateral lip by creating a cutaneous triangle flap above the white roll to correct the height of the medial lip.\textsuperscript{12} Ever since this technique has been introduced, the anatomical subunit technique has gained notable popularity.\textsuperscript{6} A recent study conducted in 2020 aimed to explore the current practice patterns in unilateral cleft lip repair among surgeons within the American Cleft Palate Association, it has concluded that up to 40\% of surgeons have changed their previous implemented techniques of cleft lip repair to the anatomical subunit approximation technique.\textsuperscript{6} Fisher’s technique of cleft lip repair has also been found to have a lesser revision rate for lip shortening, scar hypertrophy, and scar widening in comparison to the rotation advancement technique.\textsuperscript{21} However, the Fisher’s technique was found to be associated with a higher number of vermilion revisions which can be easily managed by a simple wedge excision of the vermilion.\textsuperscript{21}

As mentioned above, Millard’s technique for cleft lip repair remains one of the most implemented and taught methods for correcting the cleft lip anomaly worldwide.\textsuperscript{22} This also has been the case in in our institution. The anatomical subunit technique was introduced in the department by the senior author in 2015 and since then has been gaining popularity. The experience of this technique has not been studied in the Omani population prior to this study. Oman is a country nestled in the tip of Arabian Peninsula with a population of 5.1 million,
and land area of 309,500 km² divided into 11 provinces. It’s a country that has maintained a fine blend of traditional and modern Islamic culture in the Arab world, tradition is well preserved in the Omani population and consanguineous marriages are prevalent in the local population. Local Researches indicate that more than half (52%) of marriages are consanguineous.³ With first cousin unions being the most common type of consanguineous marriages, constituting of 39% of all marriages and 75% of consanguineous marriages.³ Consanguineous marriages are associated with a very high incidence of cleft lip and palate.³ The incidence of clefts has been found to be almost 1.5 time the normal worldwide prevalence of clefts due to cultural practice of consanguineous marriages in Oman.³

In our experience over a period of five years in the hands of a single surgeon in Oman, the outcome of cleft lip repair using the Anatomical Subunit Technique has been very satisfactory, and that can be further emphasized by the few numbers of revision surgeries needed in patients involved in our study. Excess vermilion is a recognized outcome of the anatomical subunit technique, for example, a study published by Paul. et al revealed that 37% of patients in their study group who has underwent unilateral cleft lip repair using the technique has required debulking of excess vermilion.²⁷ Furthermore, the parent’s satisfaction following surgery in our study was 82.5%. Most of the parents had well accepted the surgical repair with anatomical subunit technique and were willing to recommend the procedure to children with similar anomalies. Similarly, a study by Mazin et. al has aimed to compare the post-operative aesthetic outcomes between Fisher’s and Mohler’s technique.⁷ The comparison was done by laymen’s evaluation of the post-operative cases and has concluded superior aesthetic outcomes of Fisher’s technique.⁷

Another area we indented to compare with the international literature was the implementation of Fisher’s technique on a different spectrum of cleft lip severity. In our study, the repair was performed on both complete and incomplete cleft lips had produced identical results “[Figures 1&2]”. Thirty six out of the eighty-two patients had varying clefts of the soft and hard palate, we did not find a significant difference in the outcome of cleft lip repair done in children with isolated cleft lip in comparison to children with combined cleft lip and palate. This finding was also true with other published studies indicating that with accurate application of the technique, the outcomes will less likely be affected by the severity of the cleft lip.²¹,²²,²⁵
The high percentage of consanguinity (52%) in our study group highlight the need for targeted national campaigns involving premarital counselling regarding consanguinity especially as our study revealed that consanguineous families are more likely to have a 1st degree relative with cleft lip. Two out of the eighty-two children were confirmed as syndromic, one was a case of Trisomy 21 and the other was diagnosed as Velocardiofacial syndrome. Our syndrome related findings are also similar to the patterns found internationally, indicating Velo-Cardio-Facial Syndrome along with other previously mentioned syndromes to be commonly associated with this anomaly. The most common complication associated with anatomical subunit technique was vermilion excess, which were corrected for 6 patients in our study with simple wedge excision done at 18-24 months after the initial lip surgery.

The limitation of our study includes no comparison group, as involving cases repaired by another technique would have given a better insight on the superiority of the Anatomical Subunit technique. In addition, a more accurate temporal relationship might have been established if the study was prospective in nature. Moreover, in order to obtain more transparent results, the participation of families in the electronic survey was voluntary and anonymous, however voluntary participation commonly leads to lower response rates and some families have occasionally refused to participate in the survey for social reasons.

**Conclusion**

This is the first study in Oman to highlight the outcome and direct patients’ experience of the anatomical subunit technique for cleft lip anomaly in an Omani population. In our hands, it resulted in predictable and identical results as related to scar quality, nasal and lip symmetry in children with varying severity of the cleft lip. Revision for vermilion excess was needed in 7% of cases. The high percentage of consanguinity (52%) in our study highlight the need for more targeted national campaigns involving premarital counselling in the Omani population.

**Authors’ Contribution**

SH conceptualized and designed the study as well as supervised the work. MW, SPV, MA and MAS collected the data. BM was the patient coordinator for this study. MA analyzed the data. MW and SPV wrote the manuscript. All authors approved the final version of the manuscript.
Conflict of interest
The authors declare no conflicts of interest.

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References


Figure 1: A) Left incomplete cleft lip (pre-operative); B) Left incomplete cleft lip (post-operative).

Figure 2: A) Left complete cleft lip (pre-operative); B) Left complete cleft lip (post-operative).