Single Stage Cleft Lip and Palate Repair In Toddlers: Retrospective Review of Feasibility and Operative Experience

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Abstract: In children with cleft lip and palate (CLP), we aimed to compare a single-stage surgery group or all in one (AIO) approach with a 2-stage surgery group (2-SSG) of 18 and 12 toddlers, respectively. A retrospective review of 30 patients with CLP was conducted between 2007 and 2019. All in one procedure was performed at 12 to 24 months and 2-SSG patients had lip and primary nasal correction at 3 to 9 months, followed by palatoplasty and myringotomies at 12 to 16 months. In the AIO group, 13 (72.2%) patients had unilateral CLP, while 5 (27.8%) had bilateral CLP, which is comparable to 2-SSG who had 8 (66.7%) unilateral CLP, 3 (25%) bilateral CLP, and 1 (8.3%) incomplete CL with submucous CP. The 2-SSG had a 30 minutes longer cumulative operative time and increased blood loss that was not statistically significant (P = 0.149 and 0.219, respectively). The AIO group had a slightly longer intubation (0.67 versus 0.33 day) and pediatric intensive care unit admission duration of 1.72 versus 1.67 days, (P = 0.427, 0.927), respectively. Total hospitalization time was significantly shorter with the AIO (8 versus 10.67 days, P = 0.016). The duration of postoperative pediatric intensive care unit and need for supplemental oxygen were higher in the AIO (38.9% versus 8.3%, P = 0.064). The “AIO” approach of lip, nasal, and palate surgery from 12 to 24 months completes early surgical care in a single operation. However, based on our review, this protocol must be selective; children with comorbidities or syndromes are advised to be exempted and operated in stages.

Key Words: Age, all in one, cheiloplasty, cleft, combined, lip, palate, palatoplasty, primary repair, single stage, timing, toddler

Cleft lip and palate (CLP) is a globally frequent congenital anomaly, with a higher prevalence in certain geographical areas, which causes functional setbacks to the affected individuals. In Saudi Arabia, where this study was conducted, the prevalence of nonsyndromic orofacial clefting is marginally lower than the global mean, but the prevalence of CLP is comparable to global figures. Furthermore, isolated cleft lip (CL) was found to have a significant association with consanguinity.

Due to the impact of the CLP on an individual’s functional status, self-image, and social interaction, and the number of surgeries each patient will require in a lifetime, cleft work has been a common focus for many surgical mission programs when visiting countries with limited economic status and resources. Major referral centers such as King Faisal Specialist Hospital and Research Center in Saudi Arabia often receive high numbers of CLP patients, as well as other plastic surgical patients, which also leads to lengthy surgical waiting lists for the specialty.

In children with combined CLP, we adopted the single-stage cheiloplasty and palatoplasty surgical protocol for children aged 12 months and above. The main reasons for the use of such a protocol were as follows:

1. Lengthy waiting list.
2. As per our preferred timing protocol, the palate should not be repaired before the age of 12 months.
3. In our hands, if the child is older the lip and primary nasal correction outcomes are better. Therefore, most of these single-stage primary surgeries were performed in toddlers (12–36 months).

The aim of this study was to compare single-stage surgery with cleft lip and primary nasal deformity correction (CLNDC) in combination with palatoplasty and bilateral myringotomy-ventilation tubes (BMVTs), with the conventional 2-stage surgery where CLNDC is undertaken as the first stage and palatoplasty and BMVTs at a later date. The term “all in one” (AIO) has been used for the combined type of approach and it is quite to the point. The objective was to review a single surgeon’s experience and the feasibility of the AIO approach from a practical and technical point of view, including both unilateral and bilateral clefts.

MATERIALS AND METHODS

Study Design

This was a retrospective, single-center study that was conducted through chart reviews in the Plastic and Reconstructive Surgery section of King Faisal Specialist Hospital, Jeddah, Saudi Arabia. It
included all patients under the age of 5 years, with unilateral or bilateral CLP, who underwent surgery between 2007 and 2019 by the same surgeon. The cases included non-syndromic and syndromic CLP.

**Patient Selection**

After searching the hospital database and using all the possible ICD (International classification of diseases) codes for the period between 2007 and 2019, the study sample comprised 150 patients. All patients with isolated CL or isolated CP, those who had surgery after the age of 5, those who had either the lip, nose, palate, or all, operated on by a different surgeon, were excluded from the study. All surgeries were performed by a single plastic surgeon who had subspecialized in pediatrics. After applying the inclusion and exclusion criteria, the final sample comprised 30 patients. Patients were then categorized into 2 groups. The single-stage surgical group (SSSG) had palatoplasty and CLNDC in a single operation. The 2-stage surgical group (2-SSG) had the procedures according to the conventional staged approach. The first stage is CLNDC, which was performed between the ages of 3 and 9 months, depending on the operating room time availability and waiting list. The second stage was palatoplasty with bilateral myringotomies, which was performed between 12 and 16 months of age.

**Surgical Procedures**

Preoperatively, all patients were assessed by a general pediatrion and a pediatric anesthesiologist. One day preoperatively, hospital admission was required for all patients. Postoperatively, all SSSG patients and all postpalatoplasty (second stage surgery of the 2-SSG) were admitted to the pediatric intensive care unit (PICU) as a routine procedure.

In SSSG or the (AIO group) the lip, palate, and nose are repaired in a single surgery, including the BMVTs. The operation was performed between the ages of 12 and 24 months. The surgeon starts the operation by repairing the palate. This is followed by CLNDC.

In the 2-SSG, the first stage is the CLNDC, which is performed between 3 and 12 months of age and the second stage is palatoplasty which is performed between 12 and 16 months of age. In all cases, BMVTs were performed by an ENT surgeon.

The technique used for palatoplasty was usually the Bardach 2-flap palatoplasty with a vomerine flap for unilateral CP and 4-flap palatoplasty for bilateral CP. Regarding CL repair, modifications of Millard’s and/or Norodhoff’s techniques were often used to repair unilateral as well as bilateral CLs.

**Statistical Analysis**

Patient data (demographics, age at repair, type of procedure, surgical technique used, comorbidities, syndromes, and complications) were collected using a standardized data collection sheet. All collected data were tabulated in a spreadsheet format. Following data collection, statistical analyses were performed. All categorical variables, such as sex, comorbidities, and syndromes, are presented as numbers and percentages. Continuous variables are presented as means ± standard deviation. Nonparametric tests were used when data were skewed using the Kolmogorov–Smirnov test. Pearson chi-square or Fisher exact test was applied according to whether the expected cell frequency was smaller than 5 and was used to examine the significance of the relationship between categorical variables. An independent sample t-test was used to determine the mean significant difference between stages and characteristics of the study variables. A 2-tailed P value less than 0.05 was considered significant for all comparisons. All data were entered and analyzed using SPSS version 25 (SPSS Inc., Chicago, IL).

**RESULTS**

The total number of patients who met the inclusion criteria was 30. Eighteen patients where in the AIO group where they underwent CLNDC and palatoplasty in the same operation, including myringotomies, while 12 patients underwent staged repair (2-SSG).

The basic demographic and clinical data are shown in Supplementary Digital Content, Table 1, http://links.lww.com/SCS/D170.

Most of the patients had unilateral CLP (21/30 cases) (70%), while 8/30 (26.7%) had bilateral CLP, and only 1 patient (3.3%) had an incomplete CL with a submucous cleft palate. Four patients (13.3%) had other comorbidities, including ventricular septal defect, atrial septal defect, hepatitis B, lobar emphysema, and seizure disorder. Three (10%) were syndromic patients, 2 had Goldenhar syndrome, and 1 patient had CHARGE syndrome. Eight patients (26.7%) had respiratory-related concerns and required supplemental oxygen. Two out of the 30 patients, or more specifically 1 case from each group (6.6%) required reintubation in the PICU due to postoperative laryngeal edema and stridor. Both patients were known to have Goldenhar syndrome. One patient (3.3%) had postoperative bleeding from the palatoplasty site that required packed red blood cells replacement and no surgical intervention. The mean age for palatoplasty was 15.77 months. The mean operative time for the AIO procedure was 7 and a half hours, with an average of 9 days of hospitalization (Supplementary Digital Content, Table 2, http://links.lww.com/SCS/D171).

A comparison between the surgical groups is shown in Supplementary Digital Content, Table 3, http://links.lww.com/SCS/D172. All demographic data, including sex, age, and BMI, were comparable between the groups (P = 0.654, 0.162, and 0.5, respectively). In addition, all co-morbidities and syndromic cases were comparable between the groups with P values of 0.511 and, 0.804, respectively. In the AIO group, 13 (72.2%) patients had unilateral CLP, while 5 (27.8%) had bilateral CLP, which is comparable to the 2-SSG who had 8 (66.7%) unilateral CLP, 3 (25%) bilateral CLP, and 1 (6.7%) patient with incomplete CL with submucous CP, P value (0.460).

The 2-SSG group had a 30 minutes longer “cumulative” operative time and increased blood loss when both stages were summed, but it was not statistically significant (P = 0.149 and 0.219, respectively) (Supplementary Digital Content, Table 3, http://links.lww.com/SCS/D172). In contrast, the AIO group had a slightly longer duration of intubation (average of 0.67 versus 0.33 days) and PICU admission duration (average of 1.72 versus 1.67 days) (P value 0.427, 0.927), respectively. The total hospitalization time was significantly shorter in the AIO group (average of 8.0 versus 10.67 days, P = 0.016) (Fig. 2).

Respiratory concerns in the form of need for supplemental oxygen and lengthier PICU observation were noted in 8 (26%) patients in the entire reviewed group; however, these were significantly higher in the AIO group (38.9% versus 8.3%) (P = 0.064). The single patient who had postoperative unusual bleeding, occurred in a syndromic patient among the AIO group as well, with a (P value of 0.406), the patient responded to conservative treatment.

**DISCUSSION**

King Faisal Specialist Hospital and Research Center (a nonprofit general organization), at its Jeddah location, is a major tertiary care facility serving the western, southern, and partly the northern regions of the country. The reputation, presence of an established program, and the multidisciplinary cleft clinic make it a particularly attractive referral center for parents of children with CLP.

The plastic surgery section also receives the majority of hand and other anomalies as well various pathologic conditions, these cases also being handled by the same senior surgeon. Accordingly,
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Historically, there have been various protocols, points of view, and preferences in terms of the optimal age for palate repair, and it has certainly been a difficult question to answer. However, in most North American practices, a general consensus has been to repair the palate around the age of 1 year. The reasons postulated were largely theoretical, including better speech development and less influence on facial growth centers, compared to surgery at a later or earlier age, respectively. Another viable benefit of considering CP repair around 12 months or more is the fact that children have a much better hemodynamic status to sustain surgery in close proximity to the upper airway. In certain syndromic conditions with a retracted mandible such as Pierre Robin sequence, further delay of palatoplasty is practiced by the majority for better airway control postoperatively, a point that cannot be argued much. Having said that, surgeons’ preferences in terms of the appropriate age for a palatoplasty are influenced by the schools of thought they once belonged to. It is usually not a simple task to influence such opinions, especially for those who have been practicing for several years. As far as the CLNDC is concerned, it has been addressed at the age of 3 months or even earlier by many, largely due to request and urgency by parents, more than any solid medical reason. From our perspective, even if an opportunity permits in isolated CLN patients for earlier surgery, we in fact prefer to delay it to the age of 5 to 6 months or more, when tissues are of better size and condition for alignment and handling.

Initial reports on combined repair of the lip and palate were first made by Farina and then Davies. Kaplan published long-term results using a similar approach. We adopted the AIO approach for the following reasons:

1. Longer intubation, and surgical duration with its attendant consequences,
2. The relative delay in lip and primary nasal repair might cause a socially negative impact and embarrassment for the parents.
3. The AIO approach essentially includes 4 procedures: palatoplasty, lip repair, nasal correction, and myringotomies (Fig. 3). Our retrospective analysis involved a single surgeon’s experience over 10 years from 2010 to 2020, where the single-stage protocol was compared with the conventional staged cheiloplasty and palatoplasty. BMVTs are carried out more frequently during palatoplasty. The total number of patients was 30 children with CLP (Supplementary Digital Content, Table 1, http://links.lww.com/SCS/D170). Of these, 3 were syndromic, 21/30 were unilateral, 18/30 patients were in the AIO group, and 12/30 were in the 2-SSG. The average age at the time of surgery in the AIO group was 14.4 months (Supplementary Digital Content, Table 3, http://links.lww.com/SCS/D172). Although we would have preferred not delaying it much beyond the age of 12 months, logistic reasons do often play a significant role. In the 2-SSG, the average age for palatoplasty was 17.75 months, which is again due to the frequent delay in the CLNDC, which was carried out between 3 and 8 months in this group.

As a policy, all our patients post palatoplasty surgery need to be observed in the PICU for the first 24 hours. Some of these patients were extubated in the OR (Operating Room), while others were kept ventilated initially and extubated afterward in the PICU. Hospitalization was significantly shorter in the AIO group. Eight of the 30 patients (26%) in our review required supplemental oxygen and longer observation following extubation in the PICU. We prefer to describe this as a “respiratory concern.” Seven of these patients belonged to the AIO group. One patient from the same group, who required temporary re-intubation in the PICU, was known to be a
Goldenhar syndrome patient. However, in the 2-SSG, 1 patient as well developed laryngeal edema following his second stage surgery (palatoplasty) and required re-intubation in the PICU; this patient also had Goldenhar syndrome.

Respiratory airway “concerns” and complications are not infrequent with palatoplasty. The exact definition and differentiation of an airway or “respiratory concern” from a complication can be subjective. Some experienced anesthetists might feel comfortable exubating the child before shifting to the recovery room or PICU. Others might prefer to send patients intubated to the PICU to be extubated afterward by the PICU staff. The policy of postpalatoplasty PICU admission tends to encourage anesthetists sending patients as intubated. Therefore, when a postpalatoplasty patient is kept intubated postoperatively as per anesthetist discretion, it would be inappropriate to label it as a complication unless the patient actually deteriorated postoperatively and required a re-intubation. The duration of surgery and general anesthesia play a significant role in the need for longer intubation postsurgery, due to the quantity of anesthetic in the systemic circulation and the relatively increased soft tissue edema. Our cases in the AIO group took an average of 7.5 hours operating time, whereas the average hemoglobin of 10 g/dL was only made because the infant generally carried the advantage of more unified techniques and care, can be considered as a limitation of the study from the perspective of the child's weight at the time of CL surgery was the only significant predictive factor in terms of complications. They pointed out the need to continually validate and evaluate the dogma as the field continues to advance.

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Cleft lip repair is a major undertaking. Significant developments have taken place lately, especially in terms of nasal deformity correction and the achievement of more natural lines. Technically speaking, although magnification is a must, tissue handling and control, are much more reliable in older children. Most surgeons with our review, where the only 2 patients who required re-intubation following surgery were syndromic (Goldenhar) and the complication was not related to the group to which they belonged. Instead it was the combination of a palatal surgery in a patient with Goldenhar syndrome which is also sometimes described as cervico-mandibular dysostosis.

In terms of the limitations of our study, these can be related to a single surgeon and center, a limited number of cases, and the lack of long-term outcomes.

A single surgeon and single center experience, although it carries the advantage of more unified techniques and care, can be considered as a limitation of the study from the perspective of the applicability of our findings and recommendations to others to follow. The overall number of cases involved in this study was limited to 30, which was due to strict exclusion criteria. All revisionary cases who were treated or had a surgical stage elsewhere were excluded.

The crucial question of the effect on craniofacial growth has been previously raised, and conflicting data exist. Malek proposed early soft palate closure at 3 months, followed by simultaneous closure of the lip and hard palate at 6 months, presumably due to their theory of minimally influencing growth centers in the vomerine area. Reddy et al performed an extensive review of the value of staging the palatal surgery in cases of unilateral CLP to determine whether doing the hard palate part at a later stage would reduce the facial growth retardation, speech issues, and fistula formation, but concluded that they were unable to find reliable evidence for such a theory.

Based on some of the above studies, looking at long-term outcomes and correlating them to our practice, we believe that doing an AIO approach at the age of 12 months or above as a single long procedure, is in fact even more reassuring and encouraging. Although it is difficult to prove entirely based on our study due to the few limitations pointed out above, nevertheless we believe we are in a much better position when operating on the palate beyond the age of 12 months from the perspective of influence on growth centers, compared to other timings and protocols.

It is unfortunate that many parents are given incorrect information, usually by pediatricians, that the lip typically must be repaired by the age of 3 months. We would like to further emphasize the timing opinion regarding CL repair. It must be remembered, the common rule of thumb, “CL repair at 3 months of age” or the classic rule of 10 (age of 10 weeks, weight of 10 pounds and hemoglobin of 10 g/dL) was only made because the infant generally has better weight and hemoglobin, permitting relatively safer elective chelioplasty and healing process. Chow et al have thoroughly examined the validity of such a historic rule by reviewing the data of 1313 patients. They found that among all the 3 factors, the child’s weight at the time of CL surgery was the only significant predictive factor in terms of complications. They pointed out the need to continually validate and evaluate the dogma as the field continues to advance.
prefer using adrenaline containing infiltrations to the lip and nose, which become safer with a relatively older child (Toddler) the same can be said from the view by anesthetia. From our perspective, the overall sense of safety by the surgeon is better when operating on a toddler than an infant. In fact, there is adequate data from anesthesia and critical care literature that proves anesthetic risk does not truly decrease until after patients are aged 1 year, suggesting that surgery should be delayed until that time if anesthetic risk is in fact the most concerning factor with regards to timing. From an esthetic point of view, with our common routine of CLNDC at an age much beyond the age of 3 months and with utilization of a nasal lift or cantilever technique, we published outcomes up to 10 years of follow-up, where the lip and nose results can be observed.

It has been stated previously that the AIO approach would be ideal in countries with limited resources. The demand for complete cleft care is quite high in a vast developing country such as Saudi Arabia. It is not only the financial status of the health care service that is essential, but also the presence of a stable and dedicated team member.

It was largely logistic reasons that led us to consider the AIO in the first place; however, we now realize some of the advantages and disadvantages when such a strategy is considered. The obvious benefit of completing the initial surgical care with 1 admission is rewarding. In otherwise healthy children with CLP, with availability of other specialties and optimum postoperative care, we recommend the consideration of an AIO mission as toddlers. Based on our findings, children with associated systemic anomalies or syndromes must not be subjected to such an approach. Nevertheless, routine postoperative PICU admission care is necessary for such an AIO approach. Other exceptions to this protocol include children whose parents demand early lip surgery due to their denial of the situation and psychosocial reasons. In our experience, this also tends to occur in the more severe forms of bilateral cases, where naso-alveolar molding is not available or feasible. The consideration for an early staged repair (CLNDC followed by palatoplasty) rather than AIO should be given.

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