One-stage simultaneous cleft lip and palate repair versus two-stage repair in children with complete unilateral cleft lip and palate: a randomized controlled study

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Background
Many surgical protocols are available for the management of unilateral cleft lip and palate (UCLP). The two-stage protocol (repair of cleft lip at 3–4 months old, followed by repair of cleft palate at the age of 10–18 months) is a common practice in cleft centers, including our hospital. One-stage simultaneous repair of both cleft lip and palate has been adopted in many cleft centers with satisfactory results; the main advantages of this protocol are lower theoretical costs and less use of operative facilities. The aim of this study was to compare the two different surgical protocols in children who were operated on for UCLP.

Patients and methods
A randomized controlled trial was conducted on 32 consecutive patients with unoperated UCLP, who were allocated into two groups: group A, which consisted of 14 patients consecutively treated with one-stage simultaneous closure of the lip and hard and soft palate, and group B, which consisted of 18 patients who underwent cleft lip repair and cleft hard palate repair with a vomer flap on the first sitting, and then, repair of the remaining cleft soft palate was performed in the second sitting. The two study groups were evaluated regarding the duration of surgery, the need of intraoperative blood transfusion, postoperative complications in the form of respiratory distress, soft palate disruption, and palatal fistula occurrence, for at least 6 months following one-stage repair in group A and 6 months after soft palate repair in group B.

Results
Both groups were comparable regarding mean age at first operation (P=0.056), sex distribution (P=0.821), total duration of surgeries (P=0.363), and need for postoperative intubation (P=0.568). There was no significant difference in prevalence of postoperative palatal fistula (P=1.000) and soft palate disruption (P=0.142) between both groups.

Conclusion
Both one-stage and two-stage protocols showed comparable outcomes regarding the prevalence of postoperative palatal fistula and soft palate dehiscence.

Keywords:
cleft lip, cleft palate, complete unilateral cleft lip and palate, palatal fistula

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Introduction

Throughout cleft surgery history, there has been debate concerning the optimal timing of surgical repair. A wide range of cleft lip and palate management protocols currently exist throughout the world. The outcomes of different treatment protocols for primary management of patients with complete unilateral cleft lip and palate (UCLP) may vary considerably [1,2].

One the most common timing sequence adopted in many cleft centers (including our hospital) is the two-stage protocol of the Norwegian Center at the Riks Hospital (Oslo, Norway), which used a sequence of initial repair of the hard palate with a vomer flap and simultaneous lip repair at the age of 3–4 months. This was followed by soft palate repair at the age of 10–18 months [3].

The concept of one-stage repair is based on early repair of entire clefts of the child within the first 12 months. According to this concept, cleft lip, palate, and alveolus are repaired in one surgical session simultaneously. This approach was introduced in 1966 by Davies [4]. The one-stage procedure offers several important advantages, such as less psychosocial

trauma, low cost, and possibly an improvement in speech results because of less-scarred palatal fields and the low rate of palatal fistula [5,6].

The one-stage and two-stage treatment protocols of UCLP were chosen as reference protocols because both have been adopted in numerous comparison studies. Dental arch relationship comparison showed that the one-stage protocol and two-stage protocol were equally successful [7]. Maxillofacial growth was affected to the same degree in both treatment protocols [5,8]. Meanwhile, more favorable facial morphology was noted in the two-stage protocol [9].

The aim of this study was to compare operative and postoperative outcomes in one-stage and two-stage protocols in a sample of patients with complete UCLP.

Patients and methods

Regarding ethical considerations and consent, the ethical committee of Faculty of Medicine, Ain Shams University, approved the research protocol in September 2018 (IRB: 0006379), and informed consent was obtained from the patients' parents or guardians before including them into the study.

Study design

A single-center, prospective, randomized controlled study was conducted in a sample of patients with UCLP who were randomly allocated using a sealed envelope technique into two groups:

Group A (one-stage protocol) patients were treated consecutively with simultaneous cleft lip repair, cleft hard palate repair with a vomer flap, and cleft soft palate repair in same sitting.

Group B (two-stage protocol) patients underwent cleft lip repair and cleft hard palate repair with a vomer flap in the first sitting, and then, repair of the remaining cleft soft palate was performed in the second sitting.

Patients

This study was conducted at the Department of Pediatric Surgery, Ain Shams University Hospitals, on nonsyndromic UCLP cases treated with the two different protocols, over a 36-month period (from April 2018 till April 2021). The study included 32 consecutive patients with UCLP. Patients were excluded from the study if they were unfit for general anesthesia owing to major anomalies (e.g. cardiac anomalies and lung malformations), previously operated for cleft lip palate, syndromic infants, and infants with other deformities of the face.

Preoperative management

No presurgical orthopedic treatment was carried out. Anesthesia consultation was done for all patients. The routinely requested investigations were complete blood count, bleeding and clotting time, liver and kidney functions, chest radiograph, and echocardiography. Blood product preparation and postoperative ICU beds were arranged as per the anesthetic team request. Proper treatment of any nasal or upper respiratory tract infections was done preoperatively.

Surgical management

For group A (one-stage surgery), surgery was done according to the technique described by Hodges [2], where the lip and hard and soft palate were all closed in a single operation according to the following protocol: lip was repaired following the Millard rotation-advancement technique. For hard palate repair, an extended vomer flap with tight closure of all surgical wounds on the anterior palate was performed. Relaxing incisions along the alveoli were limited to the premolar region. Soft palate closure was done by one-cut dissection of all abnormal muscle insertions from the posterior margin of the hard palate up to the pterygoid hamuli. Primary nose correction was performed simultaneously with the one-stage closure.

For group B (two-stage surgery), the surgical techniques that were utilized were like those described by the Oslo cleft lip and palate team in Norway [10]. In the first operation, the lip was closed using the Millard technique and simultaneous hard palate closure was done using a single layer vomer flap. Then, the soft palate was closed in the second stage using the von Langenbeck technique.

All patients were operated on with the same surgical methods and by the same experienced three surgeons (the authors: A.A.A., W.A.G., and A.B.R.).

Operative outcome measures

Data about total time required for operation (time between initiation of incision and last stitch in group A, and the sum of timing of both procedures done in group B) and the need for perioperative blood transfusion were recorded.

Postoperative management and outcome measures

Postoperative follow-up started immediately after operation for any bleeding, respiratory distress, or any other complications. Plain water orally was given

after 4 h of operation. Patients were discharged on the first postoperative day, with the advice of topical and oral antibiotics, analgesia, and liquid diet with syringe (for at least for 4 weeks). Patients were also advised to come for the removal of lip stitches 7–10 days following lip repair.

All patients were followed up for at least a 6-month period after the operation to detect occurrence of palatal dehiscence and palatal fistula.

Data management and analysis

Data were collected, revised, coded, and introduced to a PC using the Statistical Package for Social Science (IBM SPSS, version 20) (Armonk, New York, USA). Nonnumerical data were presented as number and percentages, whereas numerical data with parametric distribution were presented as mean, SD, and range. The comparison between two groups with qualitative data was done by using χ^2 test and/or Fisher exact test, which was used instead of χ^2 test when the expected count in any cell was found to be less than 5 in more than 20% of cells. The comparison between two groups with quantitative data was done by using independent *t* test. The confidence interval was set to 95%, and the margin of error accepted was set to 5%. So, the *P* value was considered significant as follows: *P* value more than 0.05: nonsignificant, *P* value less than 0.05: significant, and *P* value less than 0.01: highly significant.

Results

Demographic data

A total of 32 patients with UCLP, comprising 19 males and 13 females, with age range from 2.5 to 6 months, were randomly allocated into two groups: group A, which included 14 patients, comprising eight (57.1%) males and six (42.9%) females, underwent one-stage repair. The mean±SD age at surgery was 4.21±0.70 months (range: 3.0–6.0). The mean±SD weight of the patients at the time of repair was 5.54±0.69 kg, and the patient with the least weight was 4.5 kg.

Group B included 18 patients, comprising 11 (61.1%) males and seven (38.9%) females. The mean±SD age at the first-stage operation was 3.67±0.82 months (range: 2.5–5.0), and repair of the remaining cleft soft palate was performed at the mean±SD age of 13.00±2.47 months (range: 10.0–18.0 months). The mean±SD weight was 5.11±0.96 kg in the first-stage lip repair of group B.

Both groups were comparable regarding sex, weight, and age at time of initial repair (Tables 1 and 2).

Operative data

The mean±SD operating time in group A was 192.86 ±27.99 min (range: 125–230 min), whereas in group B the mean±SD operating time was 123.89±17.68 min (range: 90–150 min) for the first-stage operation and 78.33±17.40 min (range: 50–110 min), for the second stage. The mean±SD duration of both surgeries in the two-stage protocol was 202.22±28.81 min, as shown in Table 3.

The illustrations of operative and postoperative follow-up of patients of groups A and B are shown in Figs 1 and 2, respectively.

Immediate postoperative data

Three (21.4%) patients of group A had persistently low oxygen saturation postoperatively on room air; two (14.3%) of them underwent intubation at PICU for

Table 1 Demographic data of each group

			χ^2 test		
	Group A [*n* (%)]	Group B [*n* (%)]	χ^2	*P* value	Significance
Sex					
Male	8 (57.1)	11 (61.1)	0.510	0.821	NS
Female	6 (42.9)	7 (38.9)			

NS, nonsignificant.

Table 2 Mean weight and age of patients at the time of repair and follow-up in groups A and B

	Group A		Group B		*t* test		
	Mean	SD	Mean	SD	*t*	*P* value	Significance
Weight in kg	5.54	0.69	5.11	0.96	1.39	0.174	NS
Age in months	4.21	0.70	3.67	0.82	1.99	0.056	NS
Age at 2nd stage in months (group B)			13.00	2.47			
Follow-up period in months	10.21	3.42	13.22	4.96	1.93	0.0625	NS

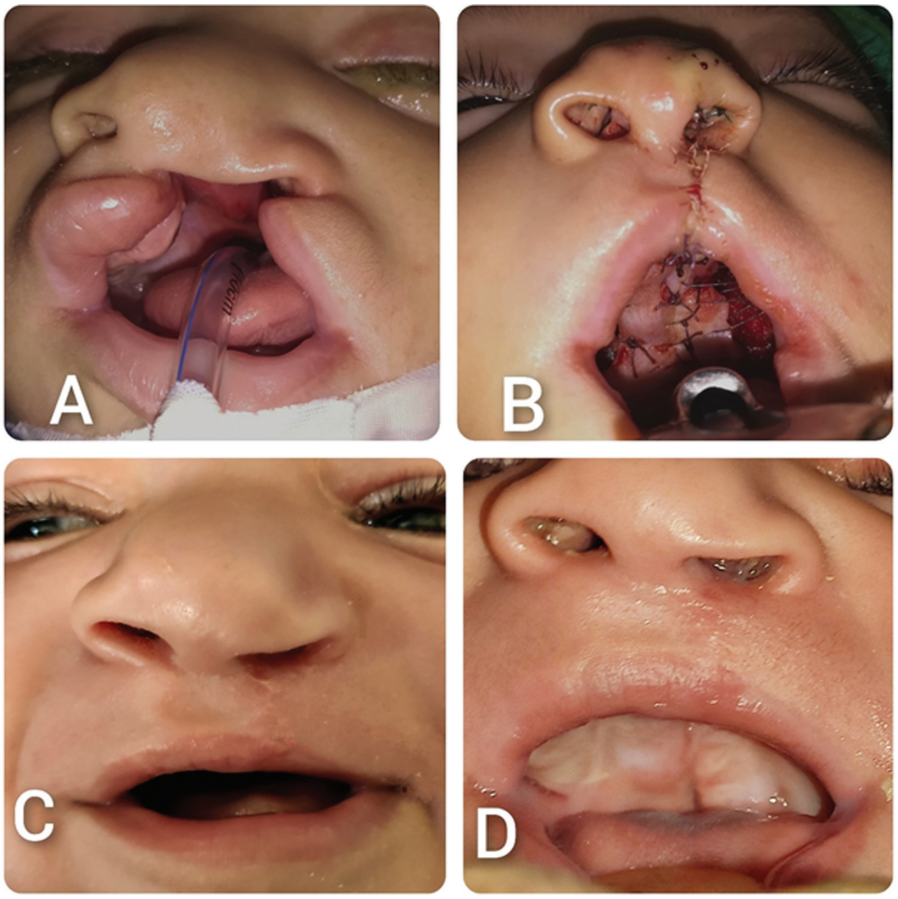
NS, nonsignificant.

Table 3 Difference in operative duration in groups A and B

	Group A		Group B		*t* test		
Total duration (min)	192.86	27.99	202.22	28.81	−0.92	0.363	NS
First-stage duration (min)			123.89	17.68			
Second-stage duration (min)			78.33	17.40			

NS, nonsignificant.

Figure 1



One-stage repair of a patient with UCLP. (a) Preoperative, (b) immediate postoperative one-stage repair, and (c, d) on review 6 months after full repair. UCLP, unilateral cleft lip and palate.

1 day postoperatively owing to airway edema, and the third one improved with noninvasive oxygen supplements. Meanwhile, two (11.1%) patients of group B had postoperative airway edema following palatal repair, where only one (5.6%) patient needed intubation for 1 day and the other patient improved with oxygen mask (Table 4).

No patients required perioperative blood transfusion, and there were no cases of intraoperative or postoperative mortality in both groups of the study.

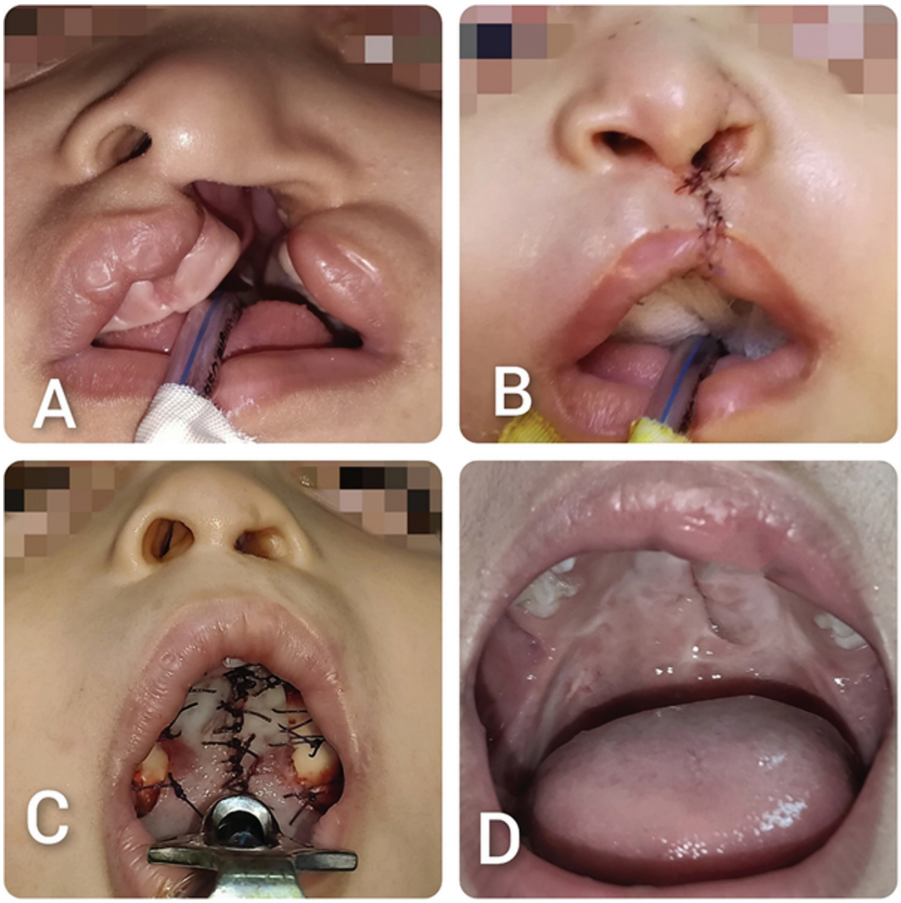
Postoperative follow-up data

All 32 patients completed more than 6 months of follow-up evaluations, and the mean±SD follow-up period for group A was 10.21±3.42 months (range: 6–18 months). Meanwhile, the mean±SD follow-up period for group B following soft palate repair was 13.22±4.96 months (range: 6–24 months) (Table 2).

Two (14.3%) patients had palatal fistula requiring closure in group A (Fig. 3a), whereas three (16.7%) patients had palatal fistulae in group B.

Four (28.6%) patients had partial soft palate dehiscence in group A (Fig. 3b), and only one (5.6%) patient had soft palate dehiscence in group B. There were no significant intergroup differences regarding the incidence of postoperative palatal fistula (P=1.000) and soft palate disruption (P=0.142) (Table 4).

Figure 2



Two-stage repair of a patient with UCLP. (a) Preoperative, (b) immediately following lip and anterior palate repair, (c) immediately following soft palate repair, and (d) on review 6 months after soft palate repair. UCLP, unilateral cleft lip and palate.

Table 4 Difference between groups A and B in postoperative complications

			Fisher exact test	
	Group A [*n* (%)]	Group B [*n* (%)]	*P* value	Significance
Airway edema				
No	11 (78.6)	16 (88.9)	0.631	NS
Yes	3 (21.4)	2 (11.1)		
ICU admission (intubation)				
No	12 (85.7)	17 (94.4)	0.568	NS
Yes	2 (14.3)	1 (5.6)		
Palatal fistula				
No	12 (85.7)	15 (83.3)	1.000	NS
Yes	2 (14.3)	3 (16.7)		
Soft palate disruption				
No	10 (71.4)	17 (94.4)	0.142	NS
Yes	4 (28.6)	1 (5.6)		

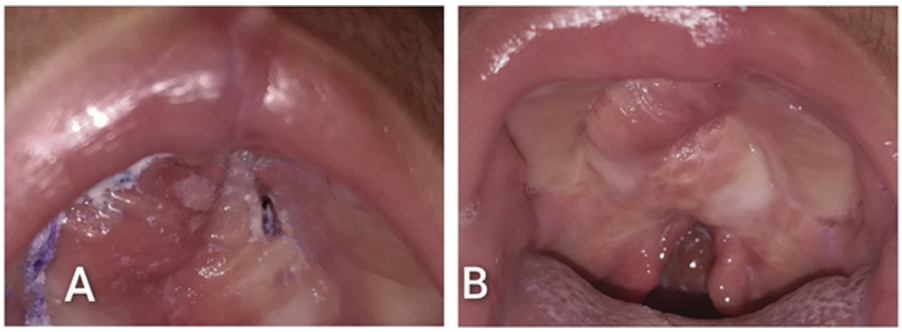
NS, nonsignificant.

Discussion

There is no universally accepted protocol for UCLP treatment. The outcome of treatments and effects of surgery seem to be mainly related to surgical protocol and technical skill of the surgeon [11].

In our hospital, we already have a long-established and successful two-stage protocol. However, the shortage of operative list availability and delay in completing the second stage of UCLP repair that affects feeding of infants led us to try the one-stage repair as an alternative approach. Before the commencement of this study, the one-stage

Figure 3



(a) Anterior palatal fistula noted 1 month after one-stage and (b) soft palatal dehiscence noted 6 months postoperatively (both in patients of group A).

protocol was introduced in our hospital with favorable initial results.

On the contrary, a debate still exists among cleft teams that early palatal closure in one-stage repair affects maxillofacial development adversely [12]. This has not been proven by long-term clinical studies [13]. Comparative retrospective studies revealed that both one-stage simultaneous repair and two-stage repair affected maxillofacial growth in patients with UCLP to the same degree [5,7].

The current study results suggested neither perioperative need of blood transfusion nor perioperative mortality in both treatment groups. These results support the general conclusion of the results of published two case series with similar outcomes, denoting the safety of the one-stage repair [2,14].

The results of the present study showed that the mean duration of one-stage surgery was shorter by almost 10 min than the mean of the sum duration of both stages of two-stage protocol. These results are in agreement with the operative findings of Guneren *et al.* [14] with one-stage protocol, where longer times were required to perform the one-stage surgery, but this elongation is shorter than the sum of the periods if the two operations had been performed separately.

The prevalence of palatal fistula in the whole study population was 15.6%, with comparable prevalence between both groups (14.3% in group A and 16.7% in group B). The prevalence of palatal fistula was similar to the mean values reported in the literature following primary palatoplasty, with values ranging from 2 to 45% [15–21]. Soft-palate dehiscence (28.6% in group A and 5.6% in group B) also showed no significant intergroup difference.

The one-stage procedure offers several important advantages over the two-stage protocol for UCLP, such as anesthesia is required only once, has a shorter period of hospitalization, is cost effective, diminishes the workload and waiting lists in our hospital, and reduces parental and infant stresses. However, we should keep in mind that performing palate repairs on small infants with the one-stage repair is challenging, as mucoperiosteal flaps are thin and need more delicate dissection in the narrower working field.

Our study was limited by the relatively small sample size and the short-term follow-up period. Thus, long-term follow-up of the patients of both study groups is recommended until complete maxillofacial growth development, to assess other treatment outcomes like maxillofacial morphology, nasolabial esthetics, and speech.

Conclusion

The present study showed comparable outcomes regarding the prevalence of postoperative soft palate dehiscence, and palatal fistula in both one-stage and two-stage treatment protocols in patients with UCLP. The one-stage simultaneous repair could be considered a safe and effective alternative procedure for the conventional two-stage protocol.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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