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The Advantages and Disadvantages of Two Stages Palatoplasty: A Systematic Review

Arya Tjipta[1]*, Hafiz Ramadhan[2], Rahmi Lubis[3]

[1]Department of Reconstructive and Aesthetic Plastic Surgery, Faculty of Medicine, Universitas Sumatera Utara, Medan, Indonesia; [2]General Practitioner, Faculty of Medicine, Universitas Sumatera Utara, Medan, Indonesia; [3]Department of Public Health, Faculty of Public Health, Universitas Sumatera Utara, Medan, Indonesia

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***Correspondence:** Arya Tjipta, Department of Reconstructive and Aesthetic Plastic Surgery, Faculty of Medicine, Universitas Sumatera Utara, Indonesia. E-mail: arya.tjipta@usu.ac.id
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Abstract

BACKGROUND: Two-stage palatoplasty involves soft palate closure–6–8 months after birth, followed by hard palate closure at the age of 12–14 years to avoid early surgical intervention of the hard palate.

AIM: This study aimed to present the advantages and disadvantages of two-stage cleft repair palatoplasty.

METHODS: This study used the literature review method to find articles using the search engine Google Scholar, Ovid, and PubMed. According to the search results, 525 articles were in accordance with the title of the study, but those in accordance with the inclusion criteria amounted to nine articles only.

RESULTS: The results of several studies in this study highlight some of the advantages and disadvantages of the two-stage palatoplasty procedure in several countries.

CONCLUSIONS: Some advantages of two-stage palatoplasty are good maxillary growth and disadvantages, such as increased velopharyngeal insufficiency.

Introduction

Labioplatoschizis or cleft lip and cleft palate/palate/palatum is a congenital abnormality that often occurs in Indonesia. The definition of cleft lip and cleft palate is the presence of a gap in the upper lip with a gap in the palate, causing a direct connection between the nose and mouth. Cleft lip disorder or cleft palate can stand alone (cleft lip only or cleft palate only) or both [1].

The crack presence in the lips and palate causes concomitant disorders, including difficulty in nutrient intake and speech function. Nutritional intake disorders caused by cleft lips or palate cause babies to find it difficult to suck or eat liquid food, which can cause other problems, namely malnutrition and difficult weight gain. The second problem is speech disorders, especially if there is a cleft palate and cleft lips which can also affect the speech patterns [1].

The treatment or management of cleft lip or palate has a variety of challenges. Since the cleft treatment requires continuous attention from baby to adulthood to achieve better final results. Cleft lip and palate abnormalities can be repaired and corrected properly. Suffered problems by patients starting from the anatomical shape of the face that is not symmetrical, nutritional problems, limited hearing and speech, susceptible to have ear infections, teeth that grow irregularly, and most importantly are the esthetic problems of facial appearance that can affect the psychological and mental development of patients [1].

Over the years, the purpose of cleft palate repair stay focused on three areas, namely, anatomical closure palatal defect, speech outcome, and minimize maxillary growth disorders [2].

Separation of the oral and nasal cavities and velopharyngeal reconstruction helps mastication eating, and prevents malnutrition [3]. Optimal development in terms of speech and restoration of articulation is very important for child development and social integration. However, prioritizing talk through early cleft palate repair can lead to growth restrictions of maxillary and often need surgical corrections [3]. Instead, prioritizing the mid-face growth by delaying hard palate repair can potentially result in speech disorders if not corrected by further surgery or speech therapy.

Surgeons distinguish their approach related how old the repair is carried out between hard palate

and soft palate, how many layers of tissue are needed to close the deformed section (defect), and the number of enough retropositioning tissues. There are still considerations of the side effects on denuded areas. Surgeons assume that this surgery has an effect on fistula, speech ability, and maxillary growth [2].

In treatment of unilateral cleft lip palate (UCLP), maxillary growth disorders in post-surgery and insufficiency of velopharyngeal have been two major concerns. One method of cleft lip repair is two stage palatoplasty which was first reported by Schweckendiek and Doz, Slaughter and Pruzansky in the 1950s [4], [5] which suggested that the closure of soft palate was carried out at the age of 6–8 months after birth, followed by hard palate closure at the age of 12–14 years to avoid early surgical intervention.

One of the procedure's characteristics of two stage palatoplasty is minimal invasion of maxillary bone until the closure of hard palate, which means that changes in procedures can affect craniofacial growth [6]. However, research on the advantages and disadvantages of this technique is still limited. The article presents the advantages and disadvantages of the two stages cleft repair procedure.

Methods

The systematic literature review was done on articles of advantages and disadvantages of two stages palatoplasty globally, freely available articles on the internet were accessed. The results of these studies were reported following preferred reporting items for systematic review and meta-analysis statement (PRISMA) 2020 guidelines.

Inclusion and exclusion

Arya Tjipta screened titles and abstracts of identified citations for potential inclusion in the review, and full texts were sought for relevant articles. Inclusion criteria for the search were published articles and electronic articles from January 2018 to March 2022 related to the advantages and disadvantages of two stages palatoplasty research globally, which were freely accessible. These included articles from primary studies and review articles (systematic review or narrative review). Studies before 2018 and non-english language articles were excluded.

Study selection

Selection of articles was performed in two stages. In the first stage, the titles and abstracts of all resources based on the inclusion criteria and search terms were screened. Selected titles and abstracts were then screened and checked whether the content potentially answered the review questions. Irrelevant abstracts were excluded and the researcher then retrieved full articles of the selected abstracts. In the second stage, full articles were screened to identify items related to the objectives of the review. Similar to the first stage, full articles were reviewed to confirm if they met the objectives of the review. The selection of articles was done using the PRISMA flow diagram as shown in Figure 1.

Search strategy and information sources

A comprehensive search to identify primary studies, reviews, and grey literature on advantages and disadvantages of two stages palatoplasty was initially conducted to capture studies published in the past 2 years. However, this period was extended till March 2022 to update the literature search before the final analysis and writing were done. The search was performed using different electronic databases (PubMed and Google Scholar). The search strategy was developed based on search terms. Boolean operators (or, and,) were used to combine the keywords (advantages, disadvantages, two stages, and palatoplasty) and related terms during the literature search.

Data extraction

Articles were excluded if they were not relevant and did not describe advantages and disadvantages of two stages palatoplasty and the objectives of the review and if the date of publication was outside January 2018–March 2022. Relevant articles were then assessed to answer the review questions. Study characteristics extracted from publications include the following author name, study design, year of publication, country of publication, target population (age), disadvantages and advantages, and limitations or gaps of the study. The results from the search were managed and extracted data from the full articles were documented in Microsoft Word.

Quality appraisal

Quality appraisal was performed on all freely accessed qualitative and quantitative studies published during the search period. Included studies were appraised for relevance. No studies were removed as a result of the quality appraisal.

Data analysis

Descriptive analysis was performed for variables relating to the year of publication and number

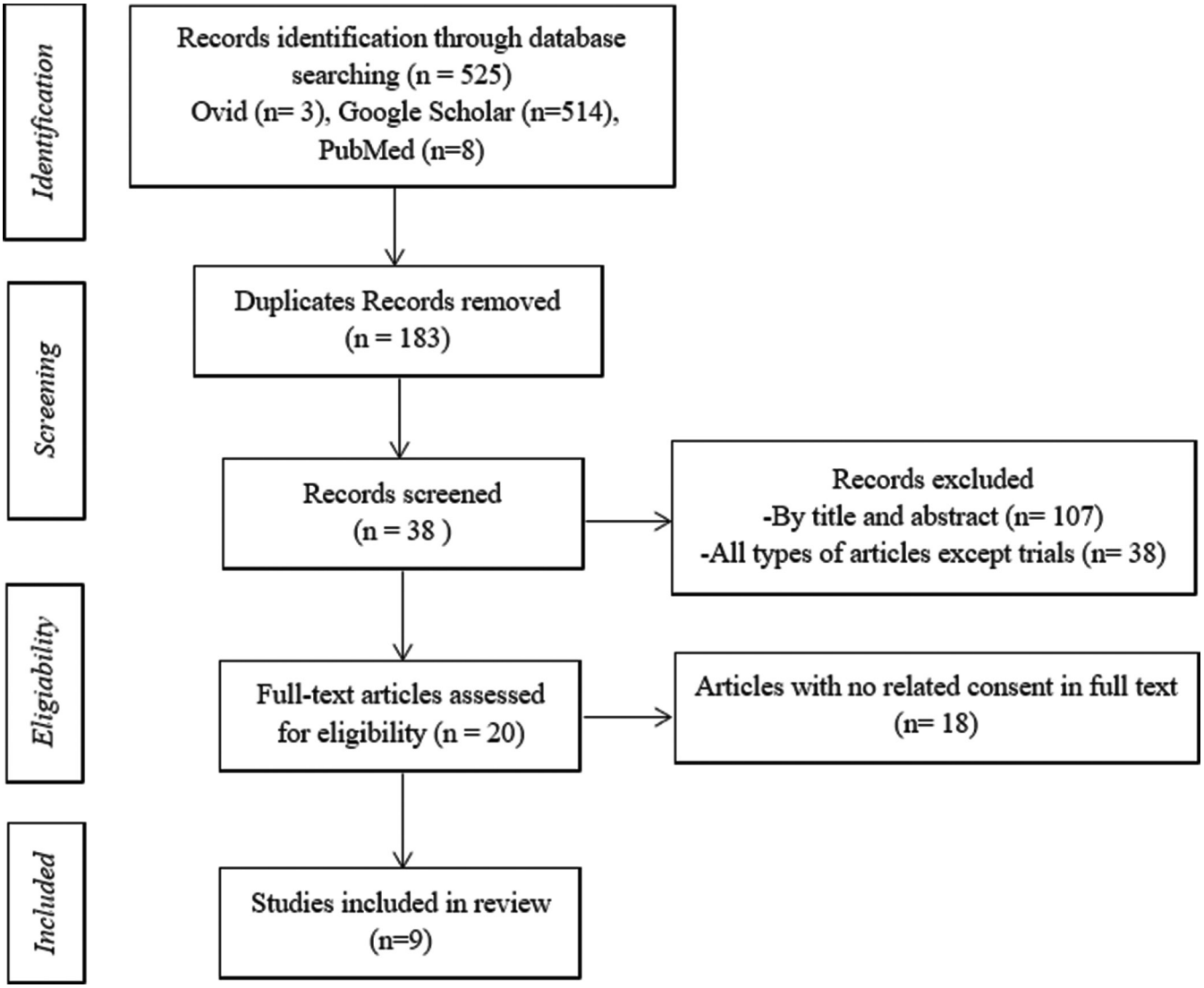

Figure 1: Details of study flow in different stages of the review

of studies, complications represented in the study, scientific article, and case report or series into which they were classified.

Results

In Table 1, some advantages that occur in patients who perform surgical procedures of two-stage palatoplasty to UCLP and asymmetric bilateral cleft lip by study design of retrospective and randomized and controlled trial. Table 1 presents information related to several scientific articles that contain about the ideal impact of surgical procedures of two-stage palatoplasty, such as starting from good jaw growth and good esthetic results to ideal tooth growth.

In Table 2, some weaknesses or similar results occur in patients who perform surgical procedures of two-stage palatoplasty to UCLP by study design of retrospective and randomized and controlled trial. Moreover, Table 2 provides information related to several scientific articles that contain about the negative impact and absence of differences from two-stage palatoplasty surgical procedures as rising the velopharyngeal insufficiency. Other data indicate that the procedure of two-stage palatoplasty has absolutely no differences compared to the other stage operations. Table 1 some scientific narratives that contain the advantages of two stage palatoplasty. Table 2 some scientific narratives that contain about the deficiency of two stage palatoplasty.

Discussion

Outcome of the cleft lip surgery must include the most favorable or ideal state of health for the patient [7]. The WHO (2018) recommends adding measures of communicative activity and participation when evaluating procedures of cleft lip palate (CLP), with the aim to

Table 1: Several Scientific Articles about Two Stages Palatoplasty

Title	Literature		Background Research		Research Design		Result
	Author	Year	Objective	Aim	Method	Sample/Variable	
Facial and nasolabial aesthetics of complete UCLP submitted to 2-stage palate repair with vomer flap [13]	Ozawa *et al*.,	2018	Patients with complete unilateral cleft lip and palate (UCLP) commonly exhibit some degree of maxillary hypoplasia as a consequence of lip and palate repair influences. Often, these patients present a class III malocclusion with anterior crossbite which can be exacerbated during the pubertal growth spurt due to differential growth between the maxilla and mandible	To evaluate the aesthetics of nasolabial appearance and facial profile of children with unilateral cleft lip and palate (UCLP) submitted to 2-stage palate repair with vomerine flap	Retrospective	Forty patients with UCLP	The nasal form and deviation was scored as good/very good in 70%, fair in 22.5%, and poor in 7.5% of the sample. The nasal–subnasal aesthetic was considered good/very good in 55%, fair in 30%, and poor in 15% of the sample. The lip vermilion border and the white part of surgical scar aesthetics were good/very good in 77.5% and 80%, fair in 17.5% for both categories, and poor in 5% and 2.5% of the cases, respectively. In all, 67.5% showed convex facial profile, 20% was straight, and 12.5% was concave profile.
Growth of palate in unilateral cleft lip and palate patients undergoing two-stage palatoplasty and orthodontic treatment [14]	Eriguchi *et al*.,	2018	A key problem with palatoplasty as primary surgery for a cleft palate is that it has to satisfy the contradictory objectives of normalizing speech while allowing maxillary growth. Therefore, much controversy has long surrounded which is the best type of surgical procedure for achieving these goals and at what age it should be performed	The purpose of this study was to investigate the long-term effects of two-stage palatoplasty on the morphology of the maxillary alveolar arch and occlusion using plaster models of the maxilla and mandible obtained from patients with unilateral complete cleft lip and palate who also underwent orthodontic treatment.	Retrospective	20 patients undergoing two-stage palatoplasty	These results suggest that two-stage palatoplasty is advantageous for jaw development.
Two-stage cleft palate closure by our treatment algorithm in complete unilateral cleft lip and palate: Results of maxillary growth at 6 years of age [15]	Mizuno *et al*.	2019	In the treatment of unilateral cleft lip, alveolus, and palate (UCLP), post-operative maxillary growth disturbance and velopharyngeal insufficiency have been two major concerns. The two-stage palatoplasty technique was first reported by Schweckendiek and Slaughter in the 1950s and involved soft palate closure at 6–8 months after birth, followed by delayed hard palate closure at age 12–14 years, in order to avoid early surgical intervention to the hard palate. This technique provided good maxillary growth, but poor post-operative velopharyngeal function	To evaluated maxillary growth outcomes of our modification of a two-stage palatoplasty algorithm (the Zurich University protocol) for the treatment of complete unilateral cleft lip, alveolus, and palate (UCLP) that allows for maxillary growth by delaying alveolar and hard palate closure with bone grafting until age 8 years or older.	Retrospective	46 consecutive patients with complete UCLP who consented to undergo treatment	Sella-nasion distance, sella-nasion-point A angle, and sella-nasion-point B angle were significantly smaller in the two-stage group than in the non-cleft group among boys but were not significantly different among girls. Profilograms showed that these smaller values reflected a smaller skull size. However, in the two-stage group, the point A-nasion-point B angle (∠ANB) was 2 standard deviations (SDs) less than the mean in 3 patients (6.5%) and 1 SD less in 10 patients (21.7%), reflecting poor maxilla-mandibular relationships. Compared with the push-back group, the two-stage group showed better maxillary growth in all measured values.
One-stage versus two-stage repair of asymmetric bilateral cleft lip: a 20-year retrospective study of clinical outcome [16]	Chung and Lo	2018	The one-stage or two-stage approaches have been a widely used technique for patients with an asymmetric bilateral cleft lip (ABCL). There is insufficient long-term outcome data between these two methods.	The purpose of this retrospective study is to compare the clinical outcome over the last 20 years.	Retrospective	A total of 95 consecutive patients were qualified for evaluation	Average follow-up was 13.1 years. 35% of the patients had a two-stage method, and 65% had a one-stage approach. All received primary nasal reconstruction. Among the satisfaction rating scores, the one-stage repair was significantly higher than two-stage reconstruction (p = 0.0001). Long-term outcomes of the two-stage patients and the unrepaired mini-microform deformities were unsatisfying in both professional and non-professional evaluators. Revision rate was higher in patients with a greaterside complete cleft lip and palate as compared with those without palatal involvement.

(Contd...)

Table 1: *(Continued)*

Title	Literature		Background Research		Research Design		Result
	Author	Year	Objective	Aim	Method	Sample/Variable	
Aesthetic outcome of the face after 2-stage palate repair for complete unilateral cleft lip and palate [17]	Feitosa	2022	Cleft lip and palate repair aims to rebuild the function of orofacial structures, decreasing the impact on language development, masticatory function and airways, as well as creating a harmonic, symmetrical nasolabial appearance with minimal scarring. There is no consensus on the best surgical technique to be adopted, but unsatisfactory results can lead to an unesthetic appearance and have negative consequences on the individual's selfesteem.	To evaluate the aesthetics of the nasolabial appearance and the facial profile of children with complete unilateral cleft lip and palate and the fistula index based on the 2-stage palatoplasty technique with vomer flap.	Retrospective	139 patients with complete unilateral cleft lip and palate submitted to the same surgical protocol and performed in a single center, Hospital for Rehabilitation of Craniofacial Anomalies (HRAC-USP)	139 children were evaluated (90 boys and 49 girls). The mean age of the population during mixed dentition photography was 6.29 years. Mean Asher-McDade Index scores ranged between 2.25 and 2.4 for all parameters. Reproducibility values ranged from moderate to substantial agreement. The incidence of palatal fistula was 21.74% and the most frequent location was in the hard palate (Pittsburgh type IV), in 36.67%. Palate function was considered adequate in 79% (n = 109/138) of individuals and 21% had speech impairment
Effect of one-stage versus two-stage palatoplasty on hypernasality and fistula formation in children with complete unilateral cleft lip and palate: a randomized controlled trial [18]	Reddy *et al*.	2018	Is one or two-stage palatoplasty more effective preventing fistula formation and hypernasality in patients with complete unilateral cleft lip and palate?	The purpose of this study was to investigate the effect of a one stage versus two stage cleft palate repair on the incidence of hypernasality and fistula formation in patients with unilateral complete cleft lip and palate.	Randomized Controlled Trial	100 patients with non-syndromic complete unilateral cleft lip and palate with a repaired cleft lip	There was no difference in fistula rates between groups. Nasalance was slightly higher in patients who had one-stage palatoplasty when compared to those that had two-stage palatoplasty, but the difference may not be clinically significant.

Table 2. Several Studies of Advantages in Two Stages Palatoplasty

Title	Literature		Background Research		Research Design		Result
	Author	Year	Objective	Aim	Method	Sample/Variable	
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 [19]	Elkasry *et al*.	2021	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 operative and postoperative outcomes in one-stage and two-stage protocols in a sample of patients with complete UCLP.	Randomized Controlled Trial	32 consecutive patients with unoperated UCLP	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.
Increased risk of velopharyngeal insufficiency in patients undergoing staged palate repair [20]	McCrary *et al*.	2020	(1) Are there differences in the incidence of VPI between singlestage or 2-stage CP repair patients? (2) Are there differences in the rate of speech surgery between groups? and (3) How does the CP repair type these patients receive contribute to their cleft-related surgical burden?	To evaluate the association of 2-stage cleft palate (CP) surgery on velopharyngeal insufficiency (VPI) incidence, speech surgeries, and cleft-related surgical burden.	Retrospective	Patients who underwent CP surgery between 2000 and 2017	A total of 1047 patients were included; 59.6% had 2-stage CP repair, 40.4% had single-stage repair. Approximately 32% of children with 2-stage CP repair were diagnosed with VPI, as opposed to 22% of single-stage patients ($p < 0.001$). Children with 2-stage CP repair were 1.8 times as likely to be diagnosed with VPI ($p < 0.001$). Speech surgery rates were similar across groups. Patients who had 2-stage repair received an average of 2.3 more cleft-related procedures, when excluding prosthesis management procedures.
Electromyographic activity of the masseter and temporal muscles in patients with nonsyndromic complete unilateral cleft lip and palate: 2-stage versus 1-stage palate repair [21]	Sabbag *et al*.	(2018)		To assess the electromyographic activity of the masseter and temporal muscles in cleft patients who underwent 1-stage palate repair versus 2-stage palate repair	Retrospective	Thirty-two patients with nonsyndromic complete unilateral cleft lip and palate	There were similar electromyographic activity of masseter and temporal muscles during mastication and at rest after 1- and 2-stage palate closure.

improve the results of ecological validity in which the results' ability of the study can be generalized to different situations or conditions [8]. Therefore, in addition to creating the procedure and timing of surgical intervention in CLP care, it is also necessary to build knowledge about what is important for the affected patient from such procedures and better guide treatment [9]. However, good care must also be considered in multidisciplinary interventions, including in surgical procedures when reporting long-term results [10], [11] especially as it has already been shown that more risky operations can lead to long-term consequences and a deterioration in the quality of life [12].

To repair this CLP, there are two approaches: One-stage and two-stage reconstruction. Each method has its own indications, advantages, and disadvantages. The choice of one method over another may depend on the clinical situation, such as the severity or type of asymmetry [3], [4] as well as the experience or preferences of the surgeon. The unique challenge is the strategy of operative decision-making based on the types of asymmetric variants to achieve symmetry. In the literature, there is still insufficient data to identify the optimal approach to evaluating long-term results for a given group and only a few have presented long-term results.

Some scientific articles have information related to the advantages of two-stage operations on this CLP. In 2018, Ozawa *et al*. explained that from 40 UCLP patients, most patients who have two-stage surgery provided adequate facial aesthetics in childhood or adolescence, especially in the subnasal part.

Article entitled the growth of palate in unilateral cleft lip and palate patients undergoing two-stage palatoplasty and orthodontic treatment provides information related to profits in the two-stage operation on CLP that of the 20 patients who underwent the operation, among them had good jaw growth.

This is similar to Mizuno *et al*., study in 2019, that 46 patients aged 6 years with a two-stage procedure of palatoplasty has better growth of the upper jaw.

Research from Brazil explained that in 2022, 139 children consisting of 90 boys and 49 girls carried out two-stage surgery with favorable results.

Research entitled The Effect of One-stage versus Two-stage Palatoplasty on Hypernasality and Fistula Formation in Children with Complete Unilateral Cleft Lip and Palate: A Randomized Controlled Trial in 2018 by Reddy *et al*., explained that there was no significant difference in one-stage and two-stage operations in cleft lip palatoplasty.

There are several disadvantages or disadvantages that can occur in the procedure of two-stage, such as increased risk of velopharyngeal insufficiency. Hillary's research in 2020 showed that improvements of cleft palate at two stages was associated with an increased risk of diagnosis of VPI. One-stage procedure for cleft palate could help lower VPI levels.

Conclusions

The conclusions of this study are some advantages of two-stage palatoplasty, such as good maxillary growth and deficiency, such as increasing velopharyngeal insufficiency.

Authors' Contributions

This paper was conceptualized and reviewed by Arya Tjipta Prananda, Hafiz Ramadhan, and Rahmi Amelia Lubis; they revised titles, abstracts, and full-text papers for appropriateness. Arya Tjipta Prananda was responsible for extracting data and data extraction was verified by Hafiz Ramadhan. The initial draft manuscript was prepared by Arya Tjipta Prananda, Hafiz Ramadhan, and Rahmi Amelia Lubis reviewed and edited the manuscript. All the authors have read and agreed to the final manuscript.

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