Correlation between Nasoalveolar Molding and Surgical, Aesthetic, Functional and Socioeconomic Outcomes Following Primary Repair Surgery: a Systematic Review

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ABSTRACT

Objectives: The authors performed a systematic review to evaluate the potential beneficial effects of the nasoalveolar molding appliance on nonsyndromic unilateral clefts of the lip and/or palate prior to primary lip repair.
Material and Methods: A literature search was performed using three electronic databases (PubMed, Embase, Web of Science) and three journals ("Cleft Palate-Craniofacial Journal", "Plastic and Reconstructive Surgery Journal" and "American Journal of Orthodontics and Dentofacial Orthopaedic") from January 1980 to April 2017. Data extraction was performed with tables treating different subjects: surgical, aesthetical, functional, socio-economical effects of nasoalveolar molding (NAM) appliances and the evolution of NAM appliances, especially three-dimensional technology.
Results: Of the 145 articles retrieved in the literature surveys, 28 were qualified for the final analysis and 20 studies were excluded because of their small sample size (less than 10 patients) and/or too long follow-up (exceeded 18 months). Four randomized controlled trials were available. Although literature allowed discussing the short-term benefits of NAM appliance and the three-dimensional technology, scientific evidence is lacking.
Conclusions: Based on the results, nasoalveolar molding appliances have positive surgical, aesthetical, functional and socio-economical effects on unilateral clefts of the lip and/or palate treatment before the primary repair surgeries. Three-dimensional technology results in a more efficient and predictable nasoalveolar molding appliance treatment. However, nasoalveolar molding appliance effect in a short term remains unclear with the available literature. Further studies that integrate three-dimensional technology in a large scale are still needed.

Keywords: cleft lip; cleft palate; orthodontic appliances; newborn infant.

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INTRODUCTION

Clefts of the lip and palate result from a fusion failure of the left and right maxillary prominences during the 6th and 12th weeks of gestation. The resultant congenital deformity leads to malformation of the upper lip, nose and alveolar and results in functional disabilities (i.e. mastication, feeding, speaking). This affects approximately 1/700 live birth in North America and the majority of clefts are unilateral and non-syndromic [1].

Two general strategies are used to close the lip and correct the nasal asymmetry. One approach involves lip and nose repair around three months of age irrespective of the size of the alveolar gap and may be followed by secondary correction of any residual deformity sometime during childhood. The second approach utilizes presurgical orthopaedic molding early after birth for approximately three months and must be performed prior to primary repair surgeries. With the latter, orthodontists and surgeons aim to provide symmetry and elongation to the deformed nasal cartilage and reduce the severity of the cleft palate prior to the initial surgical intervention [2].

Since the 1950s, McNeil [3,4] described the first intraoral presurgical orthopaedic appliance able to stimulate the tissue growth and reduce the width of the alveolar and palatal cleft. Then, clinicians have proposed several presurgical appliances to mold the alveolar arch and reduce lip and palate cleft. First, passive appliances, such as the Hotz plate, composed of a simple plate, aim to create alveolar alignment by spontaneous development of the segments without external force. Then, in the 1980s, active orthopaedics, such as the Latham appliance, are retained by surgically installed pins and delivers controlled forces to reduce the cleft gap and align the alveolar arch. Finally, in the 1990, Grayson used Matsuo et al. [5] concept and described a third category of presurgical infant appliances, semi-active, called the nasoalveolar molding (NAM) appliance. The appliance is composed of a passive alveolar plate with one (for unilateral cleft lip and/or palate [UCLP]) or two (for bilateral cleft lip and/or palate [BCLP]) nasal stents. NAM is the first appliance able to mold presurgically the alveolar arch into a predictable preferred alignment and correct the nasal cartilage at the same time [6,7]. To achieve its goal, the appliance is introduced within the first two weeks of life in order to benefit from the maximum growth potential. It must be worn full-time until the alveolar cleft is sufficiently narrowed and the lip segments are brought together. A nasal stent extending from the intraoral plate may be added to improve nasal symmetry. Primary lip repair is then performed once the effect of NAM is deemed adequate, usually after the 18 months. This surgery is more or less invasive depending on the efficiency of the NAM.

Recent reviews have been conducted to evaluate the efficacy of the presurgical orthopaedic treatment on patients with nonsyndromic unilateral clefting of the lip and/or palate. However the heterogeneity of these studies [8-12] prevents the construction of substantial evidence of the NAM effect (Table 1). Moreover, many studies evaluate the potential beneficial effects of the NAM appliance after the primary lip repair, whereas the efficiency of the appliance itself should be assessed prior to the primary lip repair (during the first 18 months).

The aim of this study was to undertaken a systematic review to compare the surgical, aesthetic, functional and socio-economical effects before the primary lip repair between patients with nonsyndromic unilateral clefting of the lip and/or palate treated either with the NAM appliance or with other presurgical appliances

Table 1. Heterogeneity between studies

Heterogeneity of studies	
Variables	**List**
Different study designs	Retrospective [36,44], prospective [32,34], randomised [58,64].
Different overall aims	Assessment of the molding of the nasal cartilage [27,44], molding of the alveolar possesses [40,42].
Mixed criteria for evaluating results	Landmarks on casts [26], cephalometry, facial photographs [43,44], photographs on cast [45,46], three-dimensional optical scanner on the cast [64], three-dimensional optical facial scanner [62,63].
Inadequate sample sizes	Small sample size [17,19], without control group [27].
Inconsistent follow-up period	Before the primary surgery (during the first 18 months), after the primary surgeries [22,23,26].
Heterogeneity of appliances	Hotz plate [41,43], Grayson NAM [40].
Variable age of the patient	When the appliance was used, when primary surgeries were performed.
Mixed level of surgeon experience [12]	

NAM = nasoalveolar molding.

or without any presurgical treatment. The objective of the study was also to highlight complications and inefficiency of the NAM appliance. The final aim of the systematic review was to introduce the evolution of the NAM appliance technique, especially ones involving three-dimensional technology that have the potential to overcome some of the inefficiencies of the current NAM appliance approach will be introduced.

MATERIAL AND METHODS

The materials and methods of the literature employed by the authors in the systematic reviews are based on comprehensive search strategies that have been discussed and standardized.

Protocol

PRISMA-P (2015) recommendations were used to methodically build this review [13].

Focus questions

The four PICO elements (population, intervention, comparison and outcome) were employed to construct the systematic reviews. The focus questions:

1. What are the outcomes of NAM prior to primary repair surgeries on the UCLP compare to the outcomes without NAM or with others appliances?
2. What are the applications of three-dimensional technology in NAM approach nowadays?

Search strategy

An electronic search of three databases (PubMed, Medline, Web of Science) was performed and included articles from January 1980 to April 2017. The research terms used to identify articles discussing NAM appliance were: MeSH terms "cleft palate" OR "cleft lip" AND non-MeSH terms: "nasoalveolar molding, "presurgical nasoalveolar molding", "presurgical orthopaedics", "presurgical orthopaedic appliance". Beside manual search in 3 journals was conducted: "Cleft Palate-Craniofacial Journal", "Plastic and Reconstructive Surgery Journal" and "American Journal of Orthodontics and Dentofacial Orthopaedic".

Type of study

The review includes all human prospective and retrospective follow-up, cohort studies, case series and randomized control studies related to NAM appliance outcomes on the UCLP.

Domain being studies

Surgical, aesthetic, functional, socio-economical effects of NAM appliances and evolution of NAM appliances, especially CAD/CAM technologies were the domain being studied in this systematic review. The outcomes were compared to results on patients treated with other presurgical appliances or patients who did not receive any presurgical treatment before the primary repair surgeries.

Inclusion criteria

Search criteria include a minimum of ten patients and a follow-up not exceeding 18 months, before the first primary repair, corresponding on the period when the facial growth is the most important. Within these studies, only the ones written in English and with clear description of treatment protocol, objective outcome measurements and proof that significant results could be attributed to use of NAM were included in this review. Studies relating to surgical, aesthetic, functional, socio-economical effects of NAM appliances and evolution of NAM appliances, especially CAD/CAM technologies were included.

Exclusion criteria

Articles that were excluded were related to the following reasons: presurgical infant orthopaedics (PSIO) different from NAM, small sample size (< 10 patients), long-term NAM efficiency reports (more than 18 months, usually after primary surgeries), bilateral clefts of the lip and/or palate and articles that lacked a control group.

Sequential search strategy

First, article titles were screened in order to exclude the impertinent studies. Then, abstracts were screened in order to exclude studies without inclusion criteria. Next, the selection of articles was performed after reviewing the "Material and Methods" and "Results" sections based on inclusion and exclusion criteria. The final stage of inclusion articles was based on evidence ratings according to the Oxford Center for Evidence-based Medicine-Level of Evidence (March 2009) [14].

Data extraction

Data extraction was performed with tables addressing different subjects: surgical, aesthetical, functional,

socio-economical effects of NAM appliances and the evolution of NAM appliances, especially three-dimensional technology. Each table includes: author, year of publication, type of study, type of cleft lip and/or palate, aim of the study, sample size, presence or not of a control group, method assessment, effect of nasal molding, effect of alveolar molding, "other" outcomes, follow-up and study limit.

Assessment of methodological quality

Two independent researchers performed the literature search. Any disagreement regarding inclusion and exclusion criteria were discussed and resolved. Recommendations proposed in the general methods for Cochrane reviews in order to reduce the risk of bias assessment in studies were followed in the selection of articles [15].

Statistical analysis

Meta-analyses were to be conducted only if there were studies of similar comparison, reporting the same outcome measures. However, the studies included revealed considerable variations in design (i.e. large diversity of no-NAM presurgical appliances, methodology and landmark used to assess the outcomes of NAM compare to other appliances).

RESULTS

Study selection and characteristics

Out of 356 articles, 145 were further evaluated (Figure 1), 48 were considered relevant according to their abstract but 20 were excluded (Table 2). Twenty-eight articles were included and classified by category in tables (Table 3 and 4): "NAM impact on primary repair surgery", "Aesthetical and functional NAM outcomes", "Efficiency of NAM compared to other appliance without nasal stents", "NAM technique improvement", "Three-dimensional technology in NAM approach", "Socio-economical impact with NAM approach", "Complications with NAM treatment". In each category author, year of publication, type of study, type of cleft lip and/or palate, aim of the study, sample size, presence or not of a control group, method assessment, effect of nasal molding, effect of alveolar molding, "other" outcomes, follow-up and study limit could be extracted.

Four types of design study were found: randomized control trials, prospective, retrospective longitudinal studies and case series. Twenty studies were excluded because of their small sample size (< 10 cleft patients) [16-21] or/and their follow-up exceeding 18 months [22-31]. Two studies relating to nasal and alveolar outcomes after PSIO treatment was excluded because the used appliance was not NAM [32,33]. Two studies were excluded because of lack of evidence [34,35].

Risk of bias within studies

Each article has been ranked according to the Oxford Center for Evidence-based Medicine-Level [14] and at two external reviewers reviewed every manuscript.

Statistical analysis

No meta-analyses could not be performed due to the heterogeneity between the studies.

NAM impact on primary repair surgeries

The purpose of the PSIO is to facilitate primary repair of the lip and reduce the secondary revision. In the present review, three articles, using control groups, were selected to illustrate the benefit of NAM. In 1998, a retrospective review on 32 patients (18 cleft patients treated with NAM and 14 treated without NAM) by Santiago et al. [36] showed that NAM associated with gingivoperiosteoplasty (GGP) decreases the number of patients who require a secondary alveolar bone graft procedure. In a recent quasi-experimental study, surgeons from the American Cleft Palate-Craniofacial Association assessed 20 photographs of cleft lip and/or palate patients [37]. They were asked to evaluate the outcome after NAM treatment, and the likelihood of these patients requiring revision surgeries. They reported that NAM seems to reduce the necessity for over corrective surgeries. The advantage of the NAM treatment is confirmed with another recent study by Broder et al. [38]. It highlighted that caregivers reported better postsurgery outcomes in the NAM group compared with no-NAM group ($P < 0.05$), particularly in relation to the appearance of the nose.

Aesthetic and functional NAM outcomes

NAM is composed of an alveolar plate and an attached nasal stent. The two parts of this device take advantage of the high plasticity of the skeletal maxillary alveolar bone and the neonatal nasal cartilage to induce an alveolar and a nasal positive remodelling. The main goal of NAM is to reduce the width of the gap between the alveolar segments. Nasal stents may be added to the palatal plate to improve the deformity of the nasal cartilages and reduce the nostril asymmetry.

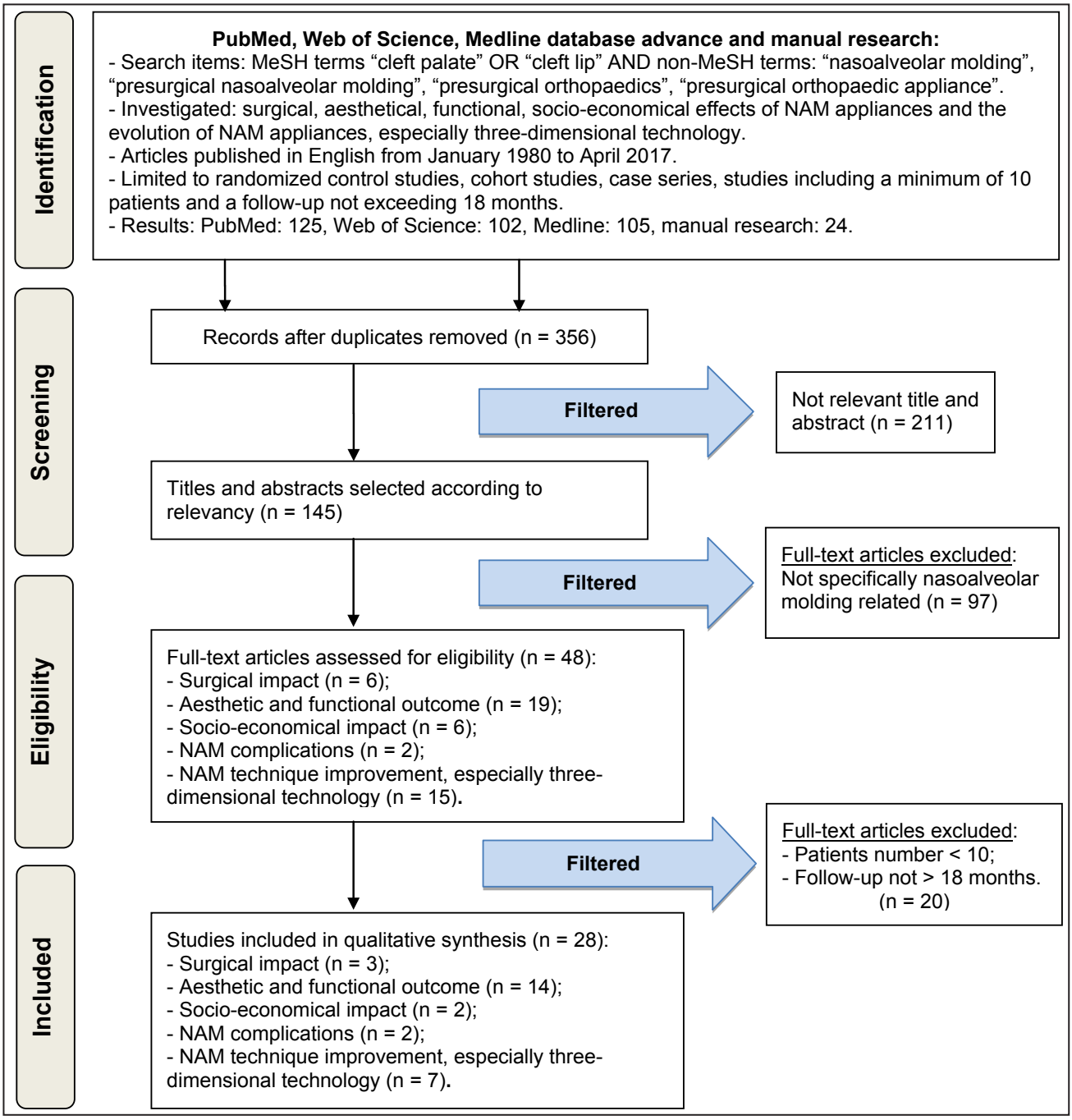

Figure 1. Flow diagram of studies selection according PRISMA guidelines.

Five prospective studies [39-43] and one retrospective study [44] compared PSIO with and without a nasal stent. All demonstrated that presurgical devices with a nasal component have the ability elongate the columella, and improve nasal asymmetry. Beside its nasal effect, three prospective studies showed that the nasal component has also a positive effect on the cleft defect [40-42]. Among the group of studies that assesses only the nasal molding, 2 studies were included. These reported an improvement of nasal symmetry and alar cartilage depression with NAM before primary surgeries [45,46].

In the present review, four publications assessed nasal and alveolar effects. All showed that the alveolar gap, nasal width and nasal asymmetry were significantly reduced at the end of the PSIO therapy, prior to the primary lip repair [47-50]. Results presented by Shetty et al. [51], highlighted that a treatment started prior to one month of age has a greater impact on the nasal symmetry than treatments started after this period. In another study by Shetty et al. [52], are comparing the patients that initiate the treatment before 1 month, between 1 and 6 months and patients that started NAM after 6 months. It showed that beneficial NAM outcomes are increased if the treatment is started as soon as possible.

Table 2. Excluded articles, classified by topic used for data extraction: surgical, aesthetical, functional, socio-economical effects of nasoalveolar molding (NAM) appliances, the improvements of NAM appliances and the three-dimensional technology (CAD/CAM)

Topic	Study	Year of publication	Study design	Cleft	Aim of the study	Sample size	Control group	Method assessment	Effect of nasal molding	Effect of alveolar molding	Other outcomes	Follow-up	Study limit	Reason for exclusion
Impacts on surgeries	Hsieh et al. [22]	2010	Retrospective study	UCLP	Effect of gingivoperiosteoplasty on facial growth	62 (NAM + GPP: 26; NAM: 40)	-	-	-	-	-	5 years	-	Follow-up exceed 18 months
	Dec et al. [23]	2013	Retrospective study	UCLP; BCLP	Assess if NAM can decrease fistula formation complication after primary repair	178	No control group	-	NAM may reduce nasolabial fistula formation	-	-	Mean: 11 years; median: 9 years	-	Follow-up exceed 18 months
	Patel et al. [24]	2015	Retrospective study	UCLP; BCLP	Assessment necessity of secondary nasal revision surgery with and without NAM	NAM: 172 UCLP, 71 BCLP; non NAM: 28 UCLP, 5BCLP	-	-	NAM: UCLP: 3%; non NAM: 21%	-	NAM treatment saves between $491 and $4893 depending on the type of cleft	5 to 14 years	-	Follow-up exceed 18 months
Aesthetic and functional impacts	Maull et al. [25]	1999	Retrospective randomized study	UCLP	Impact of NAM on long-term nasal shape	20 (presurgical nasal stent: 10; NAM: 10)	-	-	-	-	-	-	-	Follow-up exceed 18 months
	Chang et al. [26]	2010	Retrospective study	UCLP	Long-term outcome of four different techniques of nasal reconstruction	76 (NAM only: 16; NAM + rhinosplasty: 14; NAM + rhinoplasty + overcorrection: 46)	-	Two-dimensional photographs	NAM + rhino + overcorrection (20%) have best results	-	-	5 years	-	Follow-up exceed 18 months
	Liou et al. [27]	2004	Case series	UCLP	Evaluate nasal symmetry with NAM	25	-	Two-dimensional photographs	Improve nasal symmetry	-	-	3 years	No control group, small study	Follow-up exceed 18 months
	Barillas et al. [28]	2009	Retrospective study	UCLP	Long-term outcome of NAM techniques of nasal reconstruction	25 (NAM + surgical correction: 15; surgical correction only: 10)	-	Casts	Lower lateral and medial cartilage is more symmetric in the NAM group	-	-	9 years	-	Follow-up exceed 18 months
	Bennun et al. [29]	1999	Prospective study	UCLP	Compare impact on nasal symmetry	NAM: 44; presurgical orthopedics without nasal molding 47	48 healthy patients		Nasal molding permit better and permanent nasal symmetry	-	-	6 years	-	Follow-up exceed 18 months
Socio-economic aspects	Pfeifer et al. [30]	2002	Retrospective study	UCLP	Compare the cost of the financial impact of two treatment approaches	30 (group A: lip repair, nasal repair, alveolar bone graft: 14; group B: NAM, GPP, lip repair, and primary nasal repair: 16)	-		-	-	-	-	Group A cost: $22,744 Group B cost: $19,745	Follow-up exceed 18 months
	Shay et al. [31]	2015	Retrospective study	UCLP; BCLP	Compared the relative costs between cleft lip adhesion or NAM	NAM: 35; lip adhesion: 42	-	Comparison of bills	-	-	Mean costs for NAM : $3550.24 ± $667.27.Cleft adhesion costs (hospital and surgical costs): $9370.55 ± $1691.79	-	-	Follow-up exceed 18 months
	Prahl et al. [32]	2008	Prospective two-arm randomized controlled trial in parallel	UCLP	Acceptance of the treatment by mother in motherhood	NAM: 27; no-NAM: 27	-	Questionnaire	No difference between two groups	-	-	Questionnaire completed at 6, 24 and 58 weeks	-	No-NAM
	Hopkins et al. [33]	2016	Prospective study	CLP	Capture parents' lived experiences	Mother: 8; father: 4	-	-	-	-	Education and providing support can substantially improve NAM	-	-	Descriptive study
Previous technique improvments	Koya et al. [34]	2016	Prospective study with blinded measurements	UCLP	Compare traditional (Grayson) NAM with modified (Figueroa) NAM	-	-	-	-	-	-	-	No control group, only 10 patients	No control group
	Bennun et al. [35]	2006	Cases series	UCLP; BCLP	Effect of dynamic nasal bumper on nasal symmetry	UCLP: 32; BCLP: 19	-	-	Correct nasal deformity	-	Increase comfort, reduce time needed,	-	-	Method not accurate
CAD/CAM	Simanca et al. [17]	2011	Pilot study	UCLP	Measure of nasal improvement with three-dimensional photographs taken during the NAM treatment	5	None	Three-dimensional photographs	Increase columellar length on the cleft side and decrease of the nostril floor	-	Three-dimensional photograph measurement (3dMD photo system) is a reliable technique	Until 10 weeks of treatment	Small study	< 10 patients
	Braumann et al. [18]	1999	Pilot study	UCLP	Assess three-dimensional analysis system to evaluate growth rate	5	None	Three-dimensional optical scanner on the casts	-	-	Technique permit to quantify the growth rate	12 months	-	Not NAM treatment < 10 patients
	Yu et al. [19]	2011	Prospective study	UCLP	Evaluation of CAD and set of appliances made by rapid prototype technique	5	None	-	Columellar length improved	Cleft gap reduce	-	After NAM treatment	Small study	< 10 patients
	Ritschl et al. [20]	2016	Prospective	UCLP	Compare traditional NAM with CAD technology	12 (CAD NAM: 6; traditional NAM: 6)	-	-	Similar outcomes	-	Similar risk of hard and soft tissue complications	-	-	< 10 patients
	Loeffelbein et al. [21]	2015	Prospective study	UCLP	Compare two methods of planning virtual alveolar molding using CAD/CAM:	7	None	Measurement on scanned casts	-	-	Freeform method give better results (less	-	Small study	< 10 patients
	Yamada et al. [32]	2003	Prospective study	UCLP	Assess three-dimensional facial and alveolar morphology with a CAD system	15	None	Facial and alveolar forms were measured using a three-dimensional optical scanner	-	Reduction cleft gap	Make surgeries easier	Before surgical repair	-	No-NAM

CAD/CAM = computer-aided design/computer-assisted manufacture; PS = prospective study; RS = retrospective study; CS = case series; UCLP = unilateral cleft lip and/or palate; BCLP = bilateral cleft lip and/or palate; CLP = cleft lip and/or palate; GPP = gingivoperiosteoplasty.

Table 3. Included articles, classified by topic used for data extraction: impact on primary repair surgeries, on nasal and alveolar molding, nasal molding improvement with nasoalveolar molding (NAM), nasal and alveolar molding improvement with NAM and evolution of the NAM procedure

Topic	Study	Year of publication	Study design	Evidence level	Cleft	Aim of the study	Sample size	Control group	Method assessment	Effect of nasal molding	Effect of alveolar molding	Other outcomes	Follow-up
Impact on primary repair surgeries	Santiago et al. [36]	1998	Retrospective blind study	III	UCLP	Compare the need of bone graft between patients who undergo NAM + GPP and no-NAM	32 (NAM + GPP: 18)	No-NAM: 14	Clinical assessment	-	NAM group: 8 required bone graft; no-NAM group: all required bone graft	-	Before surgery
	Rubin et al. [37]	2015	Quasi-experimental study	III	UCLP	Assessment by the surgeon: necessity of secondary nasal revision surgery with and without NAM	176 of the 731 surgeons accepted to answer the survey. NAM: 10; no-NAM: 10	No-NAM: 10	Two-dimensional photographs	Necessity secondary nasal revision surgery: NAM group: 3%; no-NAM group: 21%. Not statistically significant	-	Patient with NAM cost $500 less	Before the surgery
	Broder et al. [38]	2016	Prospective non-randomized study	II	UCLP BCLP	Examines clinician and caregiver appraisals of primary cleft lip and nasal reconstruction	NAM: 62; No-NAM: 48	-	Two-dimensional photographs	Better postsurgery outcomes in the NAM group	-	-	13 months
Nasal stent impact on nasal and alveolar molding	Punga and Sharma [39]	2013	Prospective study	II	UCLP BCLP	Comparison between treatment with a presurgical appliance with and without nasal stent	20 (with nasal stents: 10; without nasal stents: 10)	-	Two-dimensional photographs	Increase the columella length with a nasal stents	-	-	Every 2 - 3 weeks until lip repair
	Monasterio et al. [40]	2013	Prospective study	II	UCLP	Compare two techniques: nasal elevator and NAM-Grayson	40 (NAM Grayson: 20; nasal elevator: 20)	-	Two-dimensional photographs casts	Two methods improved significantly the nasal asymmetry	Two methods reduced significantly the cleft width	-	3 months, (before the surgery)
	Isogawa et al. [41]	2010	Prospective study	II	UCLP	Compare effect between Hotz plate modified by adding a nasal stent and modified NAM	10 (NAM: 5; Hotz: 5)	-	Casts	Favourable effect obtains with PNAM	Favourable effect with both techniques	-	Around 130 days (4 months)
	Sasaki et al. [42]	2012	Prospective study	II	UCLP	Compare the effect between appliance with NAM and passive method with only action of alveolar plate	28 (NAM: 13; Hotz plate: 15)	Control with the symmetric nostril	Two-dimensional photographs casts	Better naris morphology in the NAM group	Cleft gap smaller in the NAM group	-	Just before the surgery and after
	Nakamura et al. [43]	2009	Prospective study	II	UCLP	Assess outcome nasal correction after NAM, compare with Hotz plate	30 (NAM: 15; Hotz: 15)		Two-dimensional photographs photographs	Better nasal shape in the NAM group	-	-	1 and 5 years post-operative
	Kozelj [44]	2007	Retrospective study	III	UCLP	Compare presurical orthopaedic without nasal stents and with stents	With nasal stents: 16; without nasal stents: 16		Two-dimensional photographs photographs	Nose was more symmetric with nasal stents	-	-	1 year after lip repair
Nasal molding improvment with NAM	López-Palacio et al. [45]	2012	Prospective study	II	UCLP	Nasal improvement with NAM	17	Non-cleft nostril	Two-dimensional photographs of casts	Improved of nasal tip projection, alar cartilage depression and	-	-	Before the primary rhinocheiloplasty (103 days),
	Gomez et al. [46]	2012	Prospective study	II	UCLP	Nasal improvement with NAM	30	-	Two-dimensional photographs of casts	Reduction of cleft columella deviation, improved columella length	-	-	Before lip surgery (146 days)
Nasal and alveolar molding improvment with NAM	Keçik and Enacar [47]	2009	Prospective study	III	UCLP	NAM effect on nasal and alveolar tissues	22	-	Two-dimensional photographs scan on cast	Reduction alar base width and the deviation of the columella	Reduction of the cleft width, arch length	-	6 months (before surgeries)
	Jaeger. et al. [48]	2007	Prospective study	III	UCLP	Evaluate nasal symmetry, gap reduction	11	-	Two-dimensional photographs	Improvement nasal symmetry and nostril shape	Cleft gap reduction	-	After NAM treatment (max 23 weeks)
	Pai et al. [49]	2005	Case-series	III	UCLP	Evaluate nasal symmetry and width	57	Non affected side	Two-dimensional photographs	Effect on nasal symmetry, height, and columella angle	-	Relapse of nostril shape in width	1 year
	Ezzat et al. [50]	2007	Prospective, blinded measurement study	II	UCLP	Evaluate improvement alveolar cleft and nose symmetry	12	-	Intra-oral and extra-oral casts	Nasal symmetry improvement	Cleft gap reduction	-	Mean 110 days
	Shetty et al. [51]	2012	Prospective study	II	UCLP	Evaluation of the NAM treatment depending on the moment when the treatment is started	45	No-NAM: 15	Two-dimensional photographs, dento-facial impression	Nasal measurements are improved with NAM group	-	-	Before NAM treatment, before surgery, at 18 months
	Shetty et al. [52]	2016	Prospective study	II	UCLP	To compare the effectiveness of NAM in infants before and after 6 months of age	150 (birth to 1 month: 50; 1 to 6 months: 50; 6 months to 1 year: 50	-	Cast landmark	Nasal height, nasal dome height, and columella height reduce with NAM.	Intersegment distance reduced	Patients who presented for treatment before 1 month of age benefited the most	1 year
Evolution of the NAM procedure	Chang et al. [58]	2014	Randomized prospective, single blind trial	II	UCLP	Compare traditional (Grayson) NAM and modified (Figueroa) NAM	30 (Grayson NAM: 15; Figueroa NAM: 15)	-	Two-dimensional photographs	Similar results in term of nasal result	-	-	Before surgery, one week after surgery and 6 month
	Liao et al. [59]	2012	Retrospective, blind study	II	UCLP	Compare traditional (Grayson) NAM and modified (Figueroa) NAM	63 (Grayson NAM: 31; Figueroa NAM: 32)	-	Two-dimensional photographs	Grayson NAM was more effective to reduce nostril width but required more corrections.	-	More ulcerations with Grayson NAM,	15 weeks

UCLP = unilateral cleft lip and/or palate; BCLP = bilateral cleft lip and/or palate; PNAM = presurgical nasoalveolar molding; n = number of patients.

Table 4. Included articles, articles concerning the three-dimensional technology (CAD-CAM)

Topic	Study	Year of publication	Study design	Evidence level	Cleft	Aim of the study	Sample size	Control group	Method assessment	Effect of nasal molding	Effect of alveolar molding	Other Outcomes	Follow-up	Study limit
Three-dimensional analysis (CAD)	Singh et al. [61]	2007	Prospective study	II	UCLP	Evaluate with three-dimensional stereophotogrammetry the facial morphology after NAM	25 (NAM + surgery: 15)	No cleft: 10	Digital stereophotogrammetry used to capture three-dimensional facial image	Three-dimensional facial morphology virtually indistinguishable from the non-cleft	-	-	37 weeks + 4 weeks after surgery	-
	Baek and Son [62]	2006	Prospective study	II	UCLP	Assess alveolar molding effect and growth with CAD	16	NA	CAD	-	Reduction cleft gap	-	2 months after cheilioplasty	No control group
	Singh et al. [63]	2005	Prospective study	III	UCLP	Evaluate three-dimensional change nasal morphology with NAM	10	NA	Three-dimensional facial image captured with digital stereophotogrammetry	Size increase on the cleft and non-cleft side. Symmetry improved	-	-	Before surgical repair	No control group
Three-dimensional analysis (CAD) and printing (CAM)	Shen et al. [53]	2015	Prospective study	II	UCLP	Evaluate effect three-dimensional printing NAM	17	None	Measured on three-dimensional computed tomography scans before and after NAM treatment	-	Alveolus became more contiguous and cleft gap was reduced	11 patients had mucosal irritation, minor mucosal ulceration, decrease cost of treatment	Post treatment (before cheiloplasty)	-
	Yu et al. [64]	2013	Randomized control study	II	UCLP	Evaluate efficiency of CAD-NAM therapy (7 - 10 pairs of appliances)	30 (CAD-NAM treated: 15)	Non-presurgical therapy: 15	Measurement on scanned cast	-	Reduce cleft gap and arch length. Decrease alveolar high	-	Average 123 days	-

UCLP = unilateral cleft lip and/or palate; BCLP = bilateral cleft lip and/or palate; CAD/CAM = computer-aided design/computer-assisted manufacture.

Reduction of the cost of presurgical treatment

Among the included studies, the one introducing three-dimensional technology report that NAM using three-dimensional technology has the potential to further decrease the cost of treatment by reducing the clinical chair time and the cost of NAM appliance [53,54].

Soft tissue, hard tissue and compliance complications

Despite the non-invasive effect of NAM therapy, some complications during and after NAM treatment can occur. One publication relating to NAM complications was included in the present review: a retrospective research [55]. According to Levy-Bercowski et al. [55], 74% of patients developed complications involving the soft tissue (mucosal ulceration, bleeding, tissue fungal infections and irritation); 7% involving the hard tissue (asymmetrical configuration of the arch) and 39% reported a lack of compliance. Among the soft and hard tissue complications, only ulcerations may be taken in account as a significant complication since the sample size is statistically small (27 patients). This suggests that improvements can be made to the traditional NAM appliance in order to reduce soft tissue problems [56].

Previous evolution of Grayson technique

Since the description of the NAM appliance by Grayson et al. [6], several modifications have been proposed. Mitsuyoshi et al. [57] reported the first modification in 2004. They suggested the use of a nasal stent constructed in cobalt-chrome wire, which is believed to enhance manual control of the force and direction of the stent by the operator [57]. Bennun and Figueroa [35] provided another modification known as dynamic presurgical nasal remodelling. Two of the selected studies assessed this last modified technique and both concluded that the Figueroa appliance (with a large nasal stent) decreases the risk of ulceration compared to the traditional (Grayson) appliance [58,59].

However, all these techniques have the potential to irritate the soft tissues, are time consuming and can discourage patients and family. After taking the impression, NAM requires manual fabrication of the appliance. The region of the cleft is filled with wax, the cast is duplicated, the plate is fabricated with heat-cured acrylic resin, and finally the surface is polished [60]. Moreover, frequent visits are needed to adjust regularly the appliance by adding sequential selective resin. Finally, all these techniques do not follow a strict and repeatable protocol and the fabrication method is totally operator dependent. They may be considered as outdated compared to innovative three-dimensional technology used in other fields of dentistry, which have embraced digital technology.

Emergence and benefit of three-dimensional analysis and printing

Rapid prototyping technology was introduced in the 1990s to medicine and dentistry. Computer-aided design (CAD) made it possible to capture digital stereo-photogrammetry with laser scanning and provided high-accurate measurements from defined landmarks. Computer-aided manufacturing (CAM) allowed printing of a sequential set of appliances obtained from the treatment design software. Three-dimensional technology for presurgical cleft treatment is widely adopted in the medical industry. Several studies have been conducted to assess this technology. Two types of studies were selected. One type assessed diagnostic efficiency with the help of CAD system, the other type evaluated the treatment effectiveness of the CAM system. Three prospective studies have been selected and all of them confirm that CAD systems are very reliable techniques to assess and quantify the nasal and alveolar improvement after presurgical treatment [61-63].

Among the studies that evaluated CAM system, one prospective study and one randomized control trial were selected [53,64]. Both showed that this method is a more efficient, more precise and more predictable technology. First, laser scanning and digital model construction simplify the NAM procedure by manufacturing a series of appliances at once, saving clinic time by reducing chair-side adjustment time. Secondly, the patient's parents are able to change the NAM appliance weekly and come to the treatment center less frequently. Third, the ability to visualize the procedure on a desktop or laptop computer can simplify communication between orthodontist and patient.

DISCUSSION

In the present review, the authors sought to answer two questions regarding the efficacy of NAM on nonsyndromic unilateral lip and/or cleft patients before primary surgeries. Does NAM appliance help presurgical orthopeadics users to better achieve cleft treatment goals, especially reduce primary

surgical needs? And, how does the three-dimensional technology improve NAM therapy?

The first question addressed the benefits of NAM on the subsequent lip repair at approximately 18 months. A study published by Santiago et al. [36] demonstrated that patients undergoing NAM followed by GGP had a reduced need of further bone grafting procedure. This trend is confirmed by surgeons and caregivers' assessments when they found that NAM treatment improve UCLP outcomes. Excluded studies (follow-up exceeding 18 months) [22,23] reinforce these findings, particularly the retrospective study conducted by Patel et al. [24]. This study showed indeed that patients with complete UCLP and BCLP treated with NAM had a lower risk of early secondary nasal revisions compare to patient without NAM. Subsequently this phenomenon explained the reduction of the cost of care with NAM treatment compare to no-NAM treatment.

This question was also related to the aesthetic and functional success of the NAM treatment. Prior studies noted that NAM significantly improves the shape of the nose and reduces the alveolar clef just before the primary repair surgeries [38-51]. Despite these positive reports, alveolar molding is still not universally utilized, and this situation is not always due to inexperience or unavailability but to scepticism regarding its efficacy. Several studies [65-70] have showed no significant effect of NAM in long-term follow-up (between 2 to 10 years old). Critical review of these has determined them to be high quality (prospective in design, large sample size, and long term follow-up). They have assessed different appliances: Latham-Millard appliance [71,72], other active appliance [65] and passive presurgical orthopaedics [66-68]. The randomized Dutchcleft series conducted in 2004 [73], 2006 [67], 2008 [74], 2009 [68] and 2015 [69,70] are some of the most cited. However, none of these latter studies assessed NAM effect in particular. Only one study, conducted by Clark et al. [75] challenged the effect of NAM by demonstrating that long-term alveolar improvement after NAM treatment was not confirmed with the three-dimensional measurements.

Another issue related to the first question is the socio-economical impact of NAM therapy because although successful treatment of the aesthetic and functional aspects of orofacial cleft anomalies is possible, it is still technically challenging, lengthy and costly. Therefore, it is important to take into consideration the economic impact of the NAM on the overall cost of the therapy and the satisfaction of parents during the presurgical orthopaedic treatment. Among included studies, Shen et al. [53] and Chen et al. [54] evaluated the cost of cleft treatment and demonstrated that the NAM appliance has the potential to decrease the overall cost of cleft treatment. Two excluded retrospective studies confirmed this trend [30,31], NAM appeared indeed as a great mean to reduce the complexity, the number of the repair surgeries and subsequently the cost of care. One of them published by Shay et al. in 2015 [31], gives interesting results and showed that the cost of a group of patients treated with NAM (mean costs $ 3550.24 ± $ 667.27) is significantly lower compare to a group of patients who received a surgical clef lip adhesion without NAM (mean costs $ 9370.55 ± $ 1691.79). No study related to parent's cooperation during NAM therapy was included. If parental compliance during NAM therapy may be compared to other presurgical orthopaedics, it is important to mention a relevant Dutchcleft prospective randomized controlled trial conducted by Prahl et al. [33]. This study compares the acceptance of presurgical orthopaedics by the mother. An analysis of questionnaires filled out by the mothers of cleft patients at 6 weeks, 24 weeks and 58 weeks showed that families were motivated to follow the orthopaedic treatment despite the increased care time required [33]. A small descriptive study by Hopkins et al. [34] that captures on parent's lived experiences, highlights that education and providing support can substantially improve NAM treatment.

A final issue that needed to be addressed was the incidence of complications that occur during NAM treatment. Two publications highlighted different sets of complications related to use of the traditional NAM appliance (vomiting, ulceration, and non-compliance). Levy-Bercowski et al. [55] offer some preventive measures to facilitate NAM fabrication and reduce complications. These include specific techniques in taking the impression, limiting the posterior limit of the device, and minimizing the thickness of the occlusal plate (2 to 3 mm) [55,76].

The second question the authors sought to answer was: does the three-dimensional technology improve NAM therapy? Three-dimensional technology is widely used in medicine [77], especially in maxillofacial surgery. CAD/CAM is able to fabricate a reproducible, accurate and individual appliance in a short period of time, which suits the goal of individualized medicine where each patient requires a specific therapeutic approach using predictive simulation systems [78]. It is now possible to produce sequential NAM devices with a three-dimensional printer from stereolithographic files obtained from predictive simulation of cleft segment manipulation.

According to Shen et al. [53], CAD [62,63] and CAM [19,53], results in a more efficient and predictable NAM treatment. Other advantages are the potential decrease in cost and time for fabrication. This technology is also more reliable and it allows for visualisation of the treatment objective, assessment of the improvement and adaptation of the appliance). Three-dimensional technology is already widely used to treat cleft deformities. A preliminary prospective study by Ritschl et al. [20] is comparing the efficiency and presence of complications between NAM made manually (n = 6) and NAM produced with three-dimensional technology (n = 6) highlighted no difference between these two methods. However; the small sample size prevents any conclusion. Prospective studies with a large sample size should be conducted to assess the real potential of three-dimensional-printed NAM devices. This innovative solution could address the shortcomings of NAM therapy and insure that NAM therapy becomes an integral part of the standard of care for unilateral cleft palate treatment.

CONCLUSIONS

According to the literature review, nasoalveolar molding appliance therapy offer positive surgical, aesthetical, functional and socio-economical effects on unilateral clefts of the lip and/or palate treatment when performed prior to primary repair surgeries. Three-dimensional technology seems to result in a more efficient and predictable nasoalveolar molding treatment. However, scientific evidence is lacking regarding the short-term effect of nasoalveolar molding appliances and the three-dimensional technology.

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