Parents’ Acceptance to Alveolar and Nasoalveolar Molding Appliances during Early Cleft Lip and Palate Care: A Call for High-Quality Research

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Abstract

AIM: Acceptance and compliance of the parents are an essential pillar in the success of pre-surgical infant orthopedics (PSIO) treatment. The aim of this systematic review is to evaluate the burden of care associated with the alveolar molding (AM) and nasoalveolar molding (NAM) appliances as experienced by the parents with unilateral complete cleft lip and palate (UCLP) infants.

METHODS: An electronic search was carried out by two reviewers in eight search engines, as well as a manual search till July 2019. Randomized controlled trials (RCTs) comparing AM/NAM appliances to controls in infants with UCLP were selected. Risk of bias was evaluated using Cochrane risk of bias assessment tool for RCTs.

RESULTS: One RCT was included in the qualitative analysis. Non-significant differences were found in the amount of mothers’ satisfaction between the intervention and control groups.

CONCLUSIONS: Insufficient low-quality evidence is available regarding the effects of AM and NAM on parents’ satisfaction and burden of care. No conclusions can be withdrawn from the existing studies. High-quality research is needed to elucidate the degree of parents’ acceptance to the molding appliances. PROSPERO registration number: CRD42016043174.

Introduction

Rationale

Cleft lip and palate (CLP) is considered the most common craniofacial anomaly in different populations [1]. Management of patients suffering from CLP can start early at infancy [2] with treatment goals of lip segments approximation, nostrils symmetry achievement, increase columellar length, and alveolar segments alignment [3]. In other words, the aim at infancy is to help the surgeons to achieve better surgical result by decreasing the severity of the cleft defect [4]. It was assumed that pre-surgical infant orthopedics (PSIO) might help in achieving these goals.

PSIO appliances were introduced in the orthodontic literature, including passive plates [5], active plates [6], pin-retained Latham’s appliance [7], and nasoalveolar molding (NAM) [3]. In 1978, Hotz et al. [8] used an intraoral plate to mold the maxillary segments before the surgical lip repair, this was only by the concept of “alveolar molding” (AM). Several years after, Grayson et al. [3] introduced the NAM concept in 1993. A nasal stent was added the intraoral plate to mold the nasal cartilage into a normal form [3] taking a further step toward achieving the PSIO goals. Both Hotz’s and Grayson’s intraoral plates were activated by addition of soft acrylic on the fitting surface at the pressure areas and grinding at the relief areas.

From all the steps of CLP management protocol, the step of PSIO is considered as the most doubtful. The researchers’ recommendations of PSIO usage were swinging between strong promoters [9], [10], [11], [12] and heavy opponents [5], [13], [14], [15], [16]. According to the latest systematic reviews [17], [18], [19], [20], NAM was considered as the most effective type of PSIO, depending on the available low-quality evidence.

Because of the handicapping nature of the infants, the success of PSIO depends mainly on the parents’ positive interaction and commitment. Several studies [16], [21], [22], [23], [24], [25], [26], [27] evaluated the parents’ satisfaction while dealing with the PSIO. They measured the amount of load on the parents, while carrying out the procedures of taking care of their child in this period. Some of those studies [25], [26] found that there is an increased burden on the parents, while others [16], [24] did not support this finding. In addition, some authors [21], [23] found that the parents were...
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Search strategy used

Results

Table 1: The electronic databases searched, the search strategies used, and the corresponding results

| Electronic database          | Date       | Search strategy used                                                                 | Results |
|------------------------------|------------|--------------------------------------------------------------------------------------|---------|
| PubMed                       | July 2019  | (Cleft lip and palate OR cleft lip OR cleft palate OR unilateral OR cleft alveolus) AND Nasoalveolar molding | 860     |
| Cochrane library             | July 2019  | OR cleft gap OR alveolar notch OR alveolar cleft AND Nasoalveolar molding              | 107     |
| LILACS                       | July 2019  | OR nasoalveolar molding OR NAM OR Pre-surgical Nasoalveolar molding OR                  | 51      |
| Scopus                       | July 2019  | nasoalveolar molding OR PNAM OR Pre-surgical appliance OR Pre-surgical appliance      | 175     |
| Wiley online Library         | July 2019  | device OR Pre-surgical orthopedics OR Pre-surgical appliance OR Nasal stents          | 23      |
| Web of Science               | July 2019  | OR nasoalveolar molding OR alveolar molding OR alveolar molding                        | 429     |
| Ovid                         | July 2019  | nasal alveolar molding OR nasal alveolar molding                                       | 8       |
| ScienceDirect                | July 2019  |                                                                                        | 237     |

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to detect attrition bias, while the sixth item is selective reporting for the reporting bias recognition. Finally, the seventh item is to evaluate any other sources of bias found by the reviewers; mostly the absence of sample size calculation was considered in this item. Three decisions to be made; either low, unclear, or high risk of bias and if a study receives a single unclear or high risk of bias the whole study takes the same evaluation.

Summary measures and synthesis of results

According to the Cochrane Handbook for systematic Reviews of interventions [29], the possible heterogeneity between the included studies was assessed in its three forms; clinical, methodological, and statistical heterogeneity. Clinical heterogeneity was assessed by comparing the demographic data of the included studies. Methodological heterogeneity was evaluated by appraisal of the followed AM/NAM protocol. In the current review, statistical heterogeneity was not assessed due to the inability to perform a meta-analysis.

Table 2: Inclusion and exclusion criteria

| Category | Inclusion criteria | Exclusion criteria |
|----------|-------------------|-------------------|
| Participants | a. Patients younger than 1 month at start of treatment. | a. Patients older than 30 days at start of treatment. |
|           | b. Infants with unilateral complete cleft lip and palate. | b. Infants with unilateral incomplete cleft lip and palate. |
|           | c. Bilateral cleft lip and palate cases. | |
| Intervention | Any technique of nasoalveolar molding | All other pre-surgical infant orthopedic devices including reversed expansion screws, pins (Latham appliance). |
| Comparator | Presence of no molding control group. | Absence of no molding control group. |
| Outcomes | Parents’ burden of care, including their ability to handle the appliance and their psychological status during NAM treatment. | Any other outcomes. |
| Study design | a. Randomized controlled trials (RCTs). | a. Prospective controlled clinical trials (CCTs). |
|           | b. Quasi randomized controlled trials (quasi-RCTs). | b. Retrospective studies. |
| Language restriction | Only studies written in English language. | |

Table 3: Data extraction sheet of the included study

| Study | Study Design and setting | Total number of patients | Patients’ age and characteristics | NAM Technique | AM or NAM (Presence of nasal stent) | Follow-up period | Types of Records | Pre-surgical records | Lip closure | Post-surgical records | Outcome |
|-------|--------------------------|--------------------------|----------------------------------|---------------|----------------------------------|-----------------|-------------------|---------------------|------------|----------------------|---------|
| 1     | Prath et al. [16] 2008   | RCT/ In three participating academic cleft palate centers in the Netherlands: Rotterdam, Amsterdam and Nijmegen. | 48 | 2 weeks/ Complete UCLP infants born at term, both parents Caucasian and fluent in the Dutch language, and trial entrance within 2 weeks after birth. | Combined treatment | No | 12 months | Questionnaire | T1*: at 6 wk. | T3: at 25 wk. | Parents Satisfaction |
| 2     | Prath et al. [16] 2008   | C | Results from this study show that infant orthopedics, with a passive plate during the first year of life, in children with a unilateral cleft lip and palate has no influence on the mothers’ satisfaction in motherhood. |

Risk of bias across studies

For assessment of publication bias, standard funnel plots and contoured enhanced funnel plots were planned to be used, only when more than ten studies included in the meta-analysis [29].

The Grades of Recommendation, Assessment, Development, and Evaluation (GRADE) [30], [31] was the tool used to assess the overall quality of evidence for each of the main outcomes. Evaluation of the methodological quality of the studies, the directness of evidence, the inconsistency, the precision of effect estimates, and the risk of publication bias were assessed using the GRADE profiler. The certainty of evidence was interpreted in four categories; very low quality, low quality, moderate quality, and high quality. Very low quality was defined as a study that is very unlikely to have an important impact on the confidence in the estimate of effect and is likely to change the estimate. Moderate quality, further research will have an important impact on the confidence in the estimate and may change it. High quality, the confidence in the estimate is high and new research is very unlikely to change this estimate.

Additional analysis

No additional analyses were performed in the presented systematic review.

Results

Study selection and characteristics

The electronic search resulted in 1917, while the manual search produced 27 studies (Figure 1). After
duplicates removal using Mendeley Desktop software (version 1.13.8), 1183 articles were subjected to screening by title and abstract. After 1178 exclusions, five studies were read in full text. As a result of full text screening, four articles [21], [22], [23], [24] were excluded with reasons (Table 4) and one study [16] met the inclusion criteria and were included in the qualitative analysis.

The included study [16] was measuring the mother’s satisfaction while dealing with the Hotz molding plate.

**Risk of bias within studies**

The Cochrane risk of bias assessment tool [28] was used for the included RCT [16] (Table 5). Seven criteria were evaluated for the included RCT. For the random sequence generation, blinding of outcome assessment, incomplete outcome data and selective reporting, the included RCT was with low risk of bias. For the allocation concealment, blinding of outcome assessment and other risks, it recorded unclear risk of bias. The overall risk of bias of the included RCT was reached to be unclear risk of bias.

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**Table 4: Excluded papers with reasons**

| Article                  | Reason for exclusion                     |
|--------------------------|-----------------------------------------|
| Sischo et al. [21] 2015  | No control group                        |
| Sischo et al. [22] 2016  | Mixed unilateral and bilateral CLP      |
| Broder et al. [23] 2016  | Mixed unilateral and bilateral CLP      |
| Hopkin et al. [24] in 2016 | No control group and retrospective study |

CLP: Cleft lip and palate.
Results of individual studies, meta-analysis, and additional analyses

The parents’ satisfaction was reported in only one RCT [16] and they found no difference between molding and no molding groups. The range of the mean scores for the individual items on the questionnaires for both groups ranged between 1.1 and 2.4. Mothers appear to be satisfied in motherhood, least satisfied with the available time for themselves, and very satisfied with hugging and walking their babies. No differences were found between groups. In the current systematic review, no meta-analyses were performed.

Risk of bias across studies

The GRADE approach for rated the available evidence as low quality for the assessed outcome (Table 6). Low quality evidence was found for parents’ satisfaction at T2 and T3.

Discussion

Controversies exist regarding the inclusion of PSIO in the followed CLP treatment protocols in the cleft centers around the world [2]. Management of infants in their 1<sup>st</sup> weeks in life is very difficult on both the parents and healthcare providers. Since that the parents are the main performers in the NAM treatment, the aim of this systematic review are to evaluate their responses with such a treatment.

A preplanned inclusion criteria were set. Studies discussed both AM and NAM appliances were included, as alveolar molding is a common step in both types. To increase the validity of this systematic review’s results, only RCTs were included. An important inclusion criterion was strictly followed, was the presence of control group for comparison.

Summary of evidence

After the systematic search, only one RCT [16] was found fitting into the inclusion criteria. This RCT made on the mothers of 48 infants with UCLP divided into 24 using Hotz plate versus 24 controls. In this study, nasal stents were not used.

In the solely found RCT [16], no differences were found between groups regarding the mother’s response to her child with and without molding. The presence of one study was insufficient to conclude the real effect of the appliances’ usage on the parents especially that, this single article did not discuss the burden of appliance posed itself, the parents’ stress and anxiety of the parents nor the effect on the fathers.

The four excluded studies [21], [22], [23], [24] evaluated the caregiver responses to the NAM appliance treatment phase. Unfortunately, these studies mixed the unilateral and bilateral cleft infants, in addition to the lack of the control group in two studies [21], [24] and that is why they were excluded from the current systematic review.

Despite of the indifferent results between the treated and the control groups in the included study [16], the excluded studies [21], [22], [23], [24] reached different conclusions. The excluded studies found a positive impact on the parents with children treated with NAM appliances. They concluded that completing the NAM treatment was often associated with positive factors such as increased empowerment, self-esteem, and bonding with their child [21], [22], in addition to more acceptable esthetic outcomes [23].

Surprisingly, none of available articles evaluated the ability of the parents to handle the

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Table 5: Assessment of risk of bias for the included RCT using Cochrane risk of bias assessment tool

| Study          | Random sequence | Allocation concealment | Blinding of participants and researchers | Blinding of outcome assessment | Incomplete outcome data | Selective reporting | Other bias | Overall risk of bias |
|----------------|------------------|------------------------|------------------------------------------|-------------------------------|-------------------------|---------------------|------------|---------------------|
| Prahl et al.   | Low              | Unclear                | Low                                      | Unclear                       | Low                     | Low                 | Unclear    | Unclear             |
| [16] 2008      |                  |                        |                                          |                               |                         |                     |            |                     |

Table 6: GRADE summary of findings table for the main outcomes of the systematic review

| Outcomes | Anticipated absolute effects (95% CI) | Risk with Alveolar and Nasoalveolar Molding | No of participants | Certainty of the evidence (GRADE) | Comments |
|----------|--------------------------------------|--------------------------------------------|--------------------|----------------------------------|----------|
| Parent Satisfaction (Parent Satisfaction) assessed with: Questionnaire Scale from: 1 to 4 follow-up: up to 58 weeks | Risk with Alveolar and Nasoalveolar Molding | No of participants | Certainty of the evidence (GRADE) | Comments |
| 48 (1 RCT) | @95-78% | LOW | Only one study found on parents satisfaction; outcome discussing only the mother’s satisfaction and found no difference between the groups |}

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<sup>1</sup> Unclear risk of bias in allocation concealment. <sup>2</sup> Only 48 patients included. <sup>3</sup> Only 28 patients were included. <sup>4</sup> High risk of bias. <sup>5</sup> Very wide confidence interval. <sup>6</sup> Confidence interval includes no effect. <sup>7</sup> Very wide confidence interval and including no effect. <sup>8</sup> Non-randomized study. <sup>9</sup> No study high risk of bias.
NAM appliances nor measuring the degree of their acceptance to the hectic appliance. All the found studies [16], [21], [22], [23], [24] evaluated the psychological side only of the parents, missing a former step of evaluating the parents’ acceptance to the appliance itself at the very beginning.

**Limitations of the available evidence**

The included RCTs were of low quality and with unclear risk of bias. No meta-analyses were performed due to the scarcely data. It seems that the degree of parents’ satisfaction was not in the interest of most of the researchers.

**Generalizability and applicability**

A knowledge gap still exists regarding the ability of the parents to handle the appliance. More high-quality studies are strongly recommended to explore this vague point.

This systematic review spots the light on a missed outcome in the literature, which is parents’ satisfaction while dealing with AM and NAM appliances. This article will motivate the researchers to assess an unexplored outcome.

**Conclusions**

Based on the available low-quality evidence, no conclusions can be withdrawn for the effect of AM or NAM on parents’ burden of care and NAM appliance acceptance. Insufficient evidence is currently available regarding the effects of AM and NAM on infants with UCLP, especially for parents’ satisfaction.

**Implications for research**

1. Further well-designed high quality RCTs are needed to evaluate parents’ satisfaction with and without NAM.
2. Post-surgical records should be available to determine the effect on the parents after lip repair.
3. The presence of control group is strongly advisable to reach true results about the burden of care.
4. Standardization of evaluation time points and the used questionnaires is important factors to allow for future pooling of data for the unevaluated outcomes.

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