

Systematic Review Cleft Lip and Palate

Impact of primary palatoplasty on the maxillomandibular sagittal relationship in patients with unilateral cleft lip and palate: a systematic review and meta-analysis

L. M. Bichara¹, R. C. Araújo¹,
C. Flores-Mir², D. Normando¹

¹Orthodontics Department, Federal University of the State of Pará, Belém, Pará, Brazil;
²School of Dentistry, University of Alberta, Edmonton, Alberta, Canada

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Abstract. The study objective was to evaluate, through a meta-analysis, the impact of primary palatoplasty on the sagittal maxillary and mandibular relationship among patients with complete unilateral cleft lip and palate (UCLP). Electronic database and hand searches were performed. Controlled clinical trials involving non-syndromic UCLP patients were included. Selected papers had to include a group of patients undergoing lip and palate repair and a group undergoing lip repair only. Data heterogeneity was demonstrated and individual means, standard deviations, and sample sizes were collected and summarized using a random effects model meta-analysis. Although six articles were selected for the systematic review, only four were included in the meta-analysis due to large discrepancies in the standard surgical protocol. Only one variable assessing the intermaxillary relationship (A point–nasion–B point; ANB), maxillary position (sella–nasion–A point; SNA), and mandibular position (sella–nasion–B point; SNB) was common among the selected studies. No significant differences in SNA and SNB were identified between patients undergoing lip surgery alone and those undergoing lip and palate surgery. Evaluation of ANB showed a small statistical standard mean difference of 0.36°. Impaired maxillary sagittal growth, observed in patients with UCLP, appears to be a basic consequence of lip surgical repair. Additional changes to the maxilla and mandible produced by palatal repair are minor. Methodologically rigorous controlled studies are needed to provide a stronger evidence-based basis for the surgical management of patients with UCLP.

Keywords: cleft lip; cleft palate; craniofacial abnormalities; surgery; oral.

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While primary repair of cleft lip and palate has not been shown to have a significant influence on mandibular morphology,¹ maxillary growth deficiencies are very common in patients with unilateral cleft lip and palate (UCLP) who have undergone an early surgical intervention.²⁻⁴

The most frequently adopted surgical protocol in the treatment of cleft considers primary palatoplasty to be the major reason for maxillary growth impairment in subjects with UCLP. This is corroborated by several studies that have compared individuals with operated UCLP vs. unoperated patients.²⁻⁸ However, some studies^{4,5} have shown that the cumulative maxillary growth disturbance attributable to lip and palate repair is not significantly worse than that determined by lip repair alone. This suggests that lip repair may be the most important factor in maxillary growth disturbance in patients with UCLP.

This comprehensive review was undertaken to review the available evidence regarding which of these surgical procedures has a greater effect on maxillary and mandibular growth. The answer to this clinically relevant question should help us to develop better treatment strategies for patients with cleft lip and palate thereby improving both functional and well-being outcomes.

Methods

The PRISMA checklist⁹ was utilized as a reporting guide.

This meta-analysis was registered at PROSPERO (the international prospective register of systematic reviews) under registration code CRD 42012003360.

For eligibility, all articles had to have compared patients with complete UCLP who had undergone lip repair followed by palate repair to patients who had undergone lip surgery and no palate repair.

Information sources

The databases used were PubMed, Cochrane Library, ScienceDirect, SciELO, and BIREME. A partial grey literature search was undertaken using Google Scholar. A hand search of the references of selected articles was also carried out to identify any article that could have been missed in the electronic database searches.

Key words 'cleft lip palate' were searched in combination with 'growth', 'surgery', and 'repair'. All references were managed using reference manager software (RefWorks) and duplicate hits were removed. Search limits were the following: controlled clinical trial, prospective or

retrospective, systematic review, meta-analysis, with human samples, no language restriction, from 1960 to the end of the search on 4 September 2013.

Study selection

The search was performed independently by two researchers (LMB and RCA). If the title and/or abstract appeared to fulfil the inclusion criteria, the article was selected for full retrieval. All abstracts had to mention patients with UCLP and a group of patients undergoing lip surgery only and a group of patients undergoing both lip and palate surgery. Additionally, all studies had to have used a lateral cephalometric analysis. Once full articles were obtained, a second selection stage was executed in which the same set of criteria was applied. Confirmation of inclusion was sought in cases where the title/abstract was misleading due to the limited description contained within them.

Data collection process

Both researchers retrieved the required information separately. The information selected was then checked jointly and any disagreements resolved. If necessary a third author (DN) was involved in the final decision.

The information retrieved from the final selection of articles included the following: author(s), year of publication, recruitment process (random, consecutive, convenience), sample size, inclusion criteria, country of origin, age at the time of surgery, error of method, sample matching (age, gender, origin, type of cleft), surgical technique, confounding factors, analysis, and the pertinent data.

Risk of bias in individual studies

A methodology checklist was applied to analyze and quantify the risk of bias in the studies included.

Summary measures

Individual means and standard deviations (SD) were collected from the lateral cephalometric measurements.

Synthesis of results

The cephalometric measurements were pooled through several meta-analyses applying a random effects model. This modelling was used because of the expected heterogeneity of the samples included. Comprehensive Meta-Analysis

software¹⁰ was used to perform the statistical analysis. All forest plots and funnel plots, as part of the meta-analysis, were also produced by the software.

Risk of bias across studies

A funnel plot was generated using the same software to verify publication bias.

Results

Study selection

Initially, 4995 articles were screened. Only 14 of them appeared to have fulfilled the inclusion criteria based on the information provided in the abstracts. Full copies of the articles were retrieved. After reading the full texts, eight articles were excluded because they did not match the primary inclusion criteria, i.e. a direct comparison of patients with UCLP undergoing surgery to the lip and palate with patients undergoing only lip repairs.¹¹⁻¹⁸ Six papers were included in the qualitative synthesis,^{2,4,5-8} however only four were included in the meta-analysis^{4,5,7,8} (Fig. 1). The studies by Mars and Houston² and Liao and Mars⁶ were excluded from the statistical analysis due to methodological issues. The first study did not record the mean age at lip repair of the sample; in the second study, the mean age at lip surgery was 7 years for patients who had undergone lip repair only and 1 year for patients who had undergone lip and palate repair. The studies included in the meta-analysis reported the following mean age at lip repair for the patients who had undergone a lip operation only (OL): 9.5 months,⁴ 44 months,⁵ 9 months,⁷ and before 24 months.⁸ The patients who had undergone lip and palate surgery (OLP) had lip surgery at a mean age of 5.5 months,⁴ 27 months,⁵ 9 months,⁷ and before 24 months.⁸ For this group, palate surgery was performed at 20 months,⁴ 54 months,⁵ 38 months,⁷ and before 36 months.⁸

Study characteristics

Sample sizes ranged from 10 subjects⁵ to 47 subjects.⁸ All patients included in the samples had UCLP. A group of patients who had undergone operations on both the lip and palate (OLP) was compared to another in which patients had undergone operations on the lip only (OL).

The authors used different surgery techniques. For lip repair, the Millard technique,^{5,6,8} Tennison technique,⁶ or rotation-advancement⁷ was used. However, patients had sometimes presented to the treatment facility with a lip repair and no clear indication of the surgery technique

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