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Review

Long-term craniofacial morphology in young adults treated for a non-syndromal UCLP: A systematic review

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Summary Minimizing mid-facial growth impairment is one of the treatment goals in cleft lip and palate surgery. As growth of the maxilla extends into young adulthood, long-term evaluation is essential to make a comprehensive assessment of a treatment protocol. There are numerous treatment approaches for cleft lip/palate surgery, and most have the characteristic distinction between either an early or a late cleft palate closure. PRISMA guidelines were applied to explore the quality of the current literature and to identify treatment factors influencing long-term cephalometric outcomes. The literature search was conducted in Pubmed, The Cochrane Library and Embase. We included studies evaluating cephalometric outcomes (SNA and ANB values on 2D cephalograms) in UCLP patients with a mean age of 16 years and older. Studies with an inadequate description of the timing of surgery were excluded. 17 studies comprising 906 patients were selected and included for critical appraisal. Treatment protocols differed considerably among the included studies and inconsistent methodology was common. Eight studies applied a one-stage procedure, 11 studies performed a two-stage reconstruction, and five studies made use of a vomer flap. Applying a multivariate model, we did not identify any treatment factors that significantly influenced growth (SNA/ANB values), except for the method of inclusion, suggesting the presence of significant selection bias within the studies. The current literature remains inadequate for evidence-based decision making and to advise parents if an early or late palate closure leads to a more favorable maxillary outgrowth. This manuscript will propose guidelines and recommended quality criteria for future studies.

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Background

Cleft lip and/or palate is the most frequently encountered congenital craniofacial malformation worldwide.[1] The main objective of cleft lip/palate treatment is to optimize speech development, aesthetic appearance, and hearing while minimizing maxillary growth impairment, which is the focus of this paper.

It is generally accepted that the iatrogenic scar tissue caused by surgical closure of a cleft interferes with maxillary growth.[2–4] Un-operated cleft lip and palate patients show better antero-posterior maxillary growth and likely have normal growth potential.[2,5] Minimizing iatrogenic mid-facial growth interference is therefore an important goal within cleft surgery. Consensus regarding the optimal surgical and orthodontic treatment protocol is however lacking. The great heterogeneity in treatment protocols and the small patient groups of previous published cohorts make it hard to differentiate between the effects of each treatment factor on mid-facial growth. Methodological discrepancies between previous studies decrease the reliability of results, complicating evidence-based decision making. Furthermore, as growth of the maxilla extends into young adulthood, long-term evaluation is essential to make a comprehensive assessment of a treatment protocol. Few cleft teams have published their long-term results and these results are often derived from small patient groups. General recommendations were provided by previous multi-center studies, but not specifically for studies evaluating cephalometric outcomes.[6]

A meta-analysis allows for the assessment of craniofacial outcomes in larger patient groups by multi-center comparison. The objective of this comprehensive review is to provide an overview of all long-term studies assessing craniofacial growth in unilateral complete cleft lip and palate (UCLP) patients and to statistically explore and determine the quality of data and reliability of their results. Furthermore, this study was conducted to perform an exploratory meta-regression to identify factors, including the timing of surgery, which could be of influence on long-term craniofacial morphology and maxillary outgrowth.

Methods

The PRISMA guidelines were utilized for the writing of this review.[7] The PRISMA checklist is included in the supplementary materials.

Clinical question

Which treatment factors influence long-term maxillary growth in patients of 17 years and older treated for a unilateral cleft lip and palate? Factors that will be assessed include: gender, timing of hard palate closure, soft palate closure, the alveolar bone graft, surgical approach (one-stage vs. two-stage palatoplasty), incidence of fistulas, incidence of pharyngoplasties.

Criteria for considering studies for this review

We included studies evaluating craniofacial morphology in UCLP patients by cephalometric analysis describing at least one of the following values: SNA and ANB. As only a few studies (n = 9) evaluated a patient group with a mean age of 18 or higher, we included all articles that had a mean age of at least 16 years old. We excluded all manuscripts in which a separate analysis of UCLP patients was not possible or where the mean cephalometric values for the studied group were not clearly described. Studies that did not define the used surgical technique or did not describe the timing of cleft surgery were also excluded from analysis. The following studies were excluded: systematic reviews, meta-analysis, level IV studies, animal studies, those that were published in languages other than English, German, French or Dutch, published before 1980, or in which patients were of non-Caucasian ethnicity.