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Maxillofacial growth and speech outcome after one-stage or two-stage palatoplasty in unilateral cleft lip and palate. A systematic review

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ABSTRACT

Background: The number of surgical procedures to repair a cleft palate may play a role in the outcome for maxillofacial growth and speech. The aim of this systematic review was to investigate the relationship between the number of surgical procedures performed to repair the cleft palate and maxillofacial growth, speech and fistula formation in non-syndromic patients with unilateral cleft lip and palate.
Material and methods: An electronic search was performed in PubMed/old MEDLINE, the Cochrane Library, EMBASE, Scopus and CINAHL databases for publications between 1960 and December 2015. Publications before 1950—journals of plastic and maxillofacial surgery—were hand searched. Additional hand searches were performed on studies mentioned in the reference lists of relevant articles. Search terms included *unilateral*, *cleft lip* and/or *palate* and *palatoplasty*. Two reviewers assessed eligibility for inclusion, extracted data, applied quality indicators and graded level of evidence.
Results: Twenty-six studies met the inclusion criteria. All were retrospective and non-randomized comparisons of one- and two-stage palatoplasty. The methodological quality of most of the studies was graded moderate to low. The outcomes concerned the comparison of one- and two-stage palatoplasty with respect to growth of the mandible, maxilla and cranial base, and speech and fistula formation.
Conclusions: Due to the lack of high-quality studies there is no conclusive evidence of a relationship between one- or two-stage palatoplasty and facial growth, speech and fistula formation in patients with unilateral cleft lip and palate.

1. Introduction

Despite considerable progress in the treatment of children with non-syndromic cleft lip and palate, there is no agreement as to the optimal timing, sequence and types of surgical procedure that yield the best result. Techniques such as the von Langenbeck (Wallace, 1987; Lindsay and Witzel, 1990), the Veau-Wardill-Kilner push-back (Wallace, 1987) and the Bardach two-flap (Bardach and Salyer, 1987; Bardach, 1995) for single-stage, and the Schweckendiek (Schweckendiek and Doz, 1978) and Delaire (Markus et al., 1993) for two-stage palatal repair were recommended. Braithwaite (1964), Kriens (1969) and Sommerlad (2003) advocated intervelar veloplasty in the soft palate by re-orientation of the levator muscle, while the Furlow Z-plasty technique was performed to improve soft palate length (Furlow, 1986).

Several earlier systematic reviews have addressed different issues regarding timing and technique of cleft palatoplasty (Nollet et al., 2005; Liao and Mars, 2006; Yang and Liao, 2010). In a systematic review on timing of hard palate repair and facial growth in 2006, the authors came to the conclusion that there is no consensus on the effect of timing on facial growth (Liao and Mars, 2006). All studies included in this review were retrospective and non-

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randomized. There was also variation in the timing of hard palate repair and inadequate assessment of outcome variables.

In 2005 a meta-analysis was published on dental arch relationships in complete unilateral cleft lip and palate based on the GOSLON yardstick for assessment of dental arch relationships (Nollet et al., 2005). The authors concluded that patients whose hard and soft palates were closed before the age of 3 had poorer GOSLON scores—indicating maxillary growth deficiency—than patients whose palates were closed at a later age.

In 2010 a systematic review was published on the effect of one-stage versus two-stage palatoplasty on maxillofacial growth (Yang and Liao, 2010). Nine studies were included, which were all retrospective and non-randomized. Timbang et al. (2014), in their systematic review, compared speech outcomes between Furlow's Z-plasty and straight-line intravelar veloplasty techniques in isolated cleft palate and unilateral cleft lip and palate. All included studies, except one, were retrospective and non-randomized. There was no statistical difference in fistula rate between Furlow and straight-line repair. The need for secondary procedures to correct velopharyngeal insufficiency in the Furlow group ranged from 0% to 6.7%, as opposed to 6.7%–19.4% in the straight-line intravelar veloplasty group. Overall, their analyses showed that straight-line repair combined with intravelar veloplasty was associated with an increased risk of a secondary surgery (1.64 times) when compared with the Furlow group.

Until now no systematic review has been published in which the results of one-stage and two-stage palatal repair are compared for different outcome variables. This systematic review was therefore carried out to examine whether one-stage or two-stage palatal repair is more beneficial for maxillofacial growth, speech and fistula rate in patients with non-syndromic unilateral complete cleft lip and palate.

2. Materials and methods

2.1. Information sources and search strategy

The search strategies were developed and databases were selected with the help of a senior librarian who specialized in health sciences. The following databases were searched: PubMed (from 1951 to 31 December 2015), Cochrane (from 1966 to 31 December 2015), EMBASE Excerpta Medica (from 1950 to 31 December 2015), SCOPUS (from 1963 to 31 December 2015), CINAHL (from 1985 to 31 December 2013).

The focus of the search was on two aspects: terms required to search for the surgical intervention of interest; and terms required to search for the congenital deformity of interest. Free text words and MeSH terms were used and individual search strings for each database were formulated, as shown in Table 1.

Publications prior to 1950—journals of plastic and maxillofacial surgery—were hand searched in. Additional hand searches were performed on studies mentioned in the reference lists of relevant articles. There was no language restriction. Grey literature (dissertations, conference abstracts) was not searched.

2.2. Eligibility criteria and study selection

The inclusion criteria for this systematic review were: study on humans; sample size of $n \geq 10$ per group; non-syndromic complete unilateral cleft lip and palate; study that compared one- and two-stage palatoplasty procedures. All reviews, isolated cleft palate studies, letters to editors and case studies and case series were excluded. No language restrictions were imposed.

Eligibility assessment of records was done based on title and abstract in an unblinded manner by two observers (AV, RR) independently. All titles and abstracts were classified as included, excluded or unclear. Inter-observer conflicts were resolved by discussion of each article to reach a consensus. In the second step, the publications classified under included or unclear were retrieved full text for further review by the two observers.

Table 1
Databases searched and search strings used.

Search engine/ database	Search terms
PubMED	(“surgery” [Subheading] OR “palate/surgery” [Mesh] OR palatoplasty) AND (unilateral [tiab] OR bilateral [tiab]) AND (“cleft palate” [MeSH Terms] OR (“cleft” [tiab] AND “palate” [tiab]) OR “cleft lip” [MeSH Terms] OR (“cleft” [tiab] AND “lip” [tiab]))
Cochrane library	(cleft lip:ti,ab,kw or cleft palate:ti,ab,kw) and (palatoplasty:ti,ab,kw or palat* surgery:ti,ab,kw or palate repair:ti,ab,kw)
EMBASE	(cleft palate/or cleft palate.mp. or cleft lip/or cleft lip.mp.) and (palatoplasty/or palatoplasty.mp.)
Scopus	(TITLE-ABS-KEY (cleft lip) AND TITLE-ABS-KEY (cleft palate) AND TITLE-ABS-KEY (unilateral) OR TITLE-ABS-KEY (bilateral) AND TITLE-ABS-KEY (palatoplasty) OR TITLE-ABS-KEY (palat* surgery) OR TITLE-ABS-KEY (palat* repair) AND TITLE-ABS-KEY (growth) OR TITLE-ABS-KEY (speech) OR TITLE-ABS-KEY (dental arch) OR TITLE-ABS-KEY (fistula))
CINAHL	(AB cleft lip OR AB cleft palate) AND (AB unilateral OR AB bilateral) AND (AB palatoplasty OR AB palat* surgery OR AB palat* repair)

2.3. Data extraction

Quantitative data extracted from each study included outcomes in relation to craniofacial form, growth of maxilla and mandible, interarch relationship, speech and fistula formation. A data extraction form was developed and piloted and finalized accordingly. Reviewers (AV, RR) independently extracted the following data from the included studies: first author, year of publication, study design, stage (one- or two-stage palatal repair), sample size, cleft type, technique of palatoplasty, timing of surgical repair, type of outcome measure, adequate and reliable measurements at follow ups, and outcomes. Disagreements were resolved by discussion between the two reviewers. If no agreement could be reached a third reviewer decided (AK).

2.4. Quality assessment and level of evidence

Two observers (AV, RR) independently evaluated the methodological quality of the included studies according to a grading system developed by the Swedish Council on Technology Assessment in Health Care, which is based on the criteria for assessing study quality from the Centre for Reviews and Disseminations (CRD) in York, UK (Deeks et al., 1996; Bondemark et al., 2007). The grades for methodological quality are listed in Table 2. The final level of evidence for each conclusion was graded according to the scale as presented in Table 3 (Bondemark et al., 2007; von Böhl et al., 2012). Conflicts, if any, between the two observers were resolved by discussion of each article.

3. Results

3.1. Study selection

The electronic search revealed a total of 5,159 citations: 2,395 from PubMed/MEDLINE, 293 from the Cochrane Library, 1,376 from EMBASE, 479 from CINAHL and 616 from SCOPUS. No additional publications were identified through hand searches. After exclusion