ORIGINAL ARTICLE

S. L. Austin C. R. Mattick P. J. Waterhouse Distraction osteogenesis versus orthognathic surgery for the treatment of maxillary hypoplasia in cleft lip and palate patients: a systematic review

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Structured Abstract

Objective – To compare the effectiveness of distraction osteogenesis to orthognathic surgery for the treatment of maxillary hypoplasia in individuals with cleft lip and palate.

Method – A systematic review of prospective randomized, quasi-randomized or controlled clinical trials. MEDLINE, EMBASE, Scopus, Web of Science, CINAHL, CENTRAL, trial registers and grey literature were searched. Hand searching of five relevant journals was completed. Two reviewers independently completed inclusion assessment. Data extraction and risk of bias assessment were completed by a single reviewer and checked by a second reviewer.

Results – Five publications all reporting different outcomes of a single randomized controlled trial are included within the review. The quality of the evidence was low with a high risk of bias. Both surgical interventions produce significant soft tissue improvement. Horizontal relapse of the maxilla was statistically significantly greater following orthognathic surgery. There was no statistically significant difference in speech and velopharyngeal function between the interventions. Maxillary distraction initially lowered social self-esteem, but this improved with time resulting in higher satisfaction with life in the long term.

Conclusions – The low quality of evidence included within the review means there is insufficient evidence to conclude whether there is a difference in effectiveness between maxillary distraction and osteotomy for the treatment of cleft-related maxillary hypoplasia. There is a need for further high-quality randomized controlled trials to allow conclusive recommendations to be made.

Key words: cleft lip and palate; distraction osteogenesis; orthognathic surgery; systematic review



Introduction

A common developmental problem in patients with cleft lip and palate is maxillary hypoplasia. Traditionally treatment for maxillary skeletal deformity was through single-stage orthognathic surgery (1). The development of maxillary distraction osteogenesis in the late 1990s has provided surgeons with an alternative method of surgical correction (2).

Management of cleft-related maxillary hypoplasia is challenging due to the risk of postsurgical relapse and potential velopharyngeal incompetence following maxillary advancement. A feature of distraction osteogenesis is that there is gradual advancement of the maxillary complex which should allow progressive adaptation of the surrounding soft tissues and palatal scar tissue. This has led to some debate as to whether distraction osteogenesis is able to produce superior post-operative outcomes compared to maxillary osteotomy. Similar to any surgical technique, distraction osteogenesis is not without its reported limitations (3,4).

The aim of this study was to complete a systematic review of the literature comparing the effectiveness of distraction osteogenesis (DO) to conventional orthognathic surgery (CO) for the treatment of maxillary hypoplasia in patients with cleft lip and palate.

Methods

Identification of studies

Electronic database searching

Electronic database searches of published literature in MEDLINE, EMBASE, Cochrane Central Register of Controlled Trials (CENTRAL), Scopus, Web of Science and Cumulative Index to Nursing and Allied Health Literature (CINAHL) were performed. A detailed search strategy was developed using a combination of controlled vocabulary and free-text terms (Table 1). The active and archived registers of the Current Controlled Trials metaRegister of Controlled Trials and the International Clinical Trials Registry Platform Search Portal were searched for relevant clinical trials. A search of OpenGrey, WorldCat Disserta-

Table 1. Search strategy for MEDLINE

OVID MEDLINE	(R) search strategy	,

- 1 Cleft lip/(MeSH)
- 2 Cleft palate/(MeSH)
- 3 (Cleftadj5lip).mp
- 4 (Cleft*adj*5*palat*).mp
- 5 (Cleft\$ adj5 oral).mp
- 6 (Cleft\$ adj5 orofacial).mp
- 7 (Cleftadj5maxill).mp
- 8 Hare-lip\$.mp
- 9 Harelip\$.mp
- 10 Cheiloschisis.mp
- 11 Palat?schi?is.mp
- 12 1 OR 2 OR 3 OR 4 OR 5 OR 7 OR 8 OR 9 OR 10 OR 11
- 13 Osteogenesis, Distraction/
- 14 (Distraction adj1 osteogen\$).mp
- 15 Osteodistraction.mp
- 16 Callus distraction.mp
- 17 Callotasis.mp
- 18 (Distraction adj5 maxill\$).mp
- 19 (Distraction adj5 midfac\$).mp
- 20 13 OR 14 OR 15 OR 16 OR 17 OR 18 OR 19
- 21 12 AND 20
- 22 Limit 21 to yr = '1996-current'

tions and Index to Theses was also undertaken to identify relevant grey literature. The database searches were undertaken between March 2012 and August 2012.

Hand searching

Hand searching of the American Journal of Orthodontics and Dentofacial Orthopedics, Cleft Palate-Craniofacial Journal, Journal of Craniofacial Surgery, Journal of Oral and Maxillofacial Surgery and Plastic and Reconstructive Surgery was completed from 1996 to July 2012 by the main reviewer.

Inclusion criteria

Participants

Individuals with moderate to severe maxillary hypoplasia due to non-syndromic cleft lip and palate suitable for correction by a conventional osteotomy. In addition they must have completed skeletal growth (16 years of age or over).

Intervention

The active intervention of distraction osteogenesis (DO).

Comparators

The control intervention of conventional orthognathic surgery (CO).

Outcome

The outcomes to be assessed were as follows:

- 1. maxillary correction achieved with the two interventions,
- 2. post-operative skeletal stability of the maxilla,
- 3. speech and velopharyngeal function,
- 4. psychological adjustment and quality of life,
- 5. outcomes relating to harms, risks and benefits.

Study design

All prospective randomized, quasi-randomized and controlled clinical trials comparing the interventions under review. Publications were excluded if they were not written in English. A time limit was applied to the search strategy to restrict it to papers published from 1996 onwards, because cleft maxillary distraction osteogenesis was not reported in the literature until 1997 (2).

Data collection and analysis

Selection of studies

Titles and abstracts were first independently assessed for inclusion by two reviewers. Full copies of all potentially relevant papers which appeared to meet the inclusion criteria or those with insufficient data to allow judgement were obtained and screened by two independent reviewers. Any disagreement regarding the eligibility of studies was resolved through discussion. Inter-rater agreement was assessed using the kappa statistic.

Data extraction

A single review author extracted the data from the included papers using a self-designed data collection form. These were checked for accuracy by a second reviewer and any disagreement was resolved by discussion.

Methodological review of clinical trials

One reviewer assessed the publications using the Cochrane Collaboration's risk of bias tool (5). The risk of bias assessments were checked by two further reviewers, and any disagreement was resolved by discussion. Each of the six domains of the risk of bias tool were rated as a high, low or unclear risk of bias.

For each publication included within the review an overall assessment of risk of bias was also made using the Cochrane Collaboration's risk of bias tools as follows:

- High overall risk of bias high risk of bias for one or more domains.
- Unclear overall risk of bias unclear risk of bias for one or more domains and low risk of bias in all other domains.
- Low overall risk of bias low risk of bias for all domains.

Data synthesis

Meta-analysis was precluded because the number of included studies was insufficient and all the publications reported different outcome measures. Therefore narrative synthesis was undertaken.

Personal contact

Study authors were contacted by e-mail to seek clarification when necessary. Unfortunately these emails were not always responded to, in which case the published information available was utilized.

Results Description of studies

The search identified 1260 references and once all duplicates were removed this left 485 references (Fig. 1). After assessment of the titles and abstracts, 14 full-text articles were reviewed for more detailed evaluation. Of these, five publications (6–10) met all the inclusion criteria (Table 2) and nine publications (11–19) were excluded (Table 3). The inter-rater agreement for the selection of studies for inclusion within this review was calculated using the kappa statistic



Fig. 1. PRISMA diagram showing the destination of studies included in the review.

as 0.833 (95% CI 0.744–0.921) and can therefore be considered to be very good.

The publications included within this systematic review are all randomized controlled trials conducted in Hong Kong. Examination of the papers appears to indicate that all of the five included publications relate to a single trial, with each paper reporting a different outcome measure. The authors were contacted to seek clarification on this matter, but no reply was received.

The participants included within the publications were all patients with cleft lip and palate aged over 16 years with moderate maxillary hypoplasia requiring 4–10 mm of maxillary advancement. All the publications consisted of a control group receiving a standardized Le Fort 1 maxillary osteotomy with titanium miniplate fixation. The intervention group had Le Fort 1 osteotomy cuts made, the maxilla was fully mobilized and an internal bone–borne maxillary distractor was placed.

The outcomes reported in the publications relate to short-term (6) and long-term maxillary stability and relapse (7), clinical morbidities (6), speech and velopharyngeal function (8), soft and hard tissue changes (9) and psychological status (10). The findings of the included publications are summarized in Table 5.

Assessment of risk of bias

The reporting of the methodology conflicted between the five included publications. Therefore

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Publication	Method	Participants	Interventions	Outcomes
Cheung et al. (6)	RCT	29 CLP patients ≥16 years with moderate maxillary hypoplasia.	Active: Internal DO Control: CO	Relapse of maxilla. Clinical morbidities. Assessed up to 1 year post-op.
Chua et al. (7)	RCT	47 CLP patients ≥16 years with moderate maxillary hypoplasia.	Active: Internal DO Control: CO	Relapse of maxilla. Maxillary incisor angulation. Assessed up to 5 years post-op.
Chua et al. (8)	RCT	47 CLP patients ≥ 16 years with moderate maxillary hypoplasia.	Active: Internal DO Control: CO	Velopharyngeal function (nasoendoscopy). Hypernasality, hyponasality and nasal emissions (perceptual speech assessment). Nasalance assessment (nasometer). Assessed up to 2 years post-op.
Chua and Cheung (9)	RCT	47 CLP patients ≥16 years with moderate maxillary hypoplasia.	Active: Internal DO Control: CO	Hard and soft tissue changes and ratios. Changes in lip thickness, nasolabial angle and nasal projection. Assessed up to 2 years post-op.
Chua et al. (10)	RCT	30 CLP patients ≥16 years with moderate maxillary hypoplasia.	Active: Internal DO Control: CO	Social avoidance and distress scale. Cultural-free self-esteem inventory. Satisfaction with life scale. Assessed up to 2 years post-op.

CLP, cleft lip and palate; DO, distraction osteogenesis; CO, conventional orthognathic surgery.

Table 3. Excluded studies

Study	Reason for exclusion
Baek et al. (11)	Not a prospective CCT or RCT.
Chanchareonsook	RCT, but does not fulfil the inclusion
et al. (12)	criteria.
Cheung et al. (13)	Not a prospective CCT or RCT.
Cheung and Chua (14)	RCT, but does not fulfil the inclusion
	criteria.
Daimaruya et al. (15)	Not a prospective CCT or RCT.
Harada et al. (16)	Not a prospective CCT or RCT.
Harada et al. (17)	Not a prospective CCT or RCT.
Kumar et al. (18)	Not a prospective CCT or RCT.
Tateishi et al. (19)	Not a prospective CCT or RCT.

RCT, randomized controlled trial; CCT, controlled clinical trial.

a risk of bias assessment was completed for each of the publications separately, according to the information presented in each article (Table 4). All the publications included within this review had some degree of methodological defect leading to a high overall risk of bias.

Effect of intervention

Skeletal relapse

Two of the papers assessed short-term (6) and long-term (7) relapse of the maxilla by comparing the vertical and horizontal movements of microscrews placed at A point and P point (mesial root apex of upper first molar) relative to horizontal and vertical reference lines on serial lateral cephalograms.

The publication reviewing short-term relapse of 29 participants up to 1 year following surgical intervention concluded that the maxillary distraction group experienced further forward and downward movement of A point and P point at each post-operative time period. Conversely in the osteotomy group the data indicates a backward and upward movement of A point and P point at every follow-up period, consistent with relapse of the maxilla (6).

The paper reviewing long-term relapse at the 5-year review period found that following maxillary distraction, the mean horizontal change of the maxilla at A point was an overall forward

	Random number sequence	Allocation concealment	Blinding	Free of incomplete outcome data	Selective outcome reporting	Other risk of bias	Overall risk of bias
Cheung et al. (6)	Low	High	High	High	Unclear	Unclear	HIGH
Chua et al. (7)	Low	High	High	High	Unclear	Unclear	HIGH
Chua et al. (8)	Low	Unclear	Low	High	High	Unclear	HIGH
Chua and Cheung (9)	Low	High	High	High	High	Low	HIGH
Chua et al. (10)	Low	High	Low	Unclear	Unclear	Unclear	HIGH

Table 4. Risk of bias summary

movement of 2.27 mm (32.3%) and 2.51 mm (35.5%) of forward movement at P point. In comparison, 5 years following maxillary osteotomy the group experienced 2.53 mm (36.9%) of backward relapse at A point and 2.45 mm (35.5%) of relapse at P point. Comparison of the two interventions found horizontal relapse of the maxilla at both A and P points was greater by a statistically significant amount in the CO group compared to the DO group 5 years post-operatively (7).

At 5 years post-operatively relapse into a class III malocclusion occurred in 3 of the 25 osteotomy patients. This compared to 1 of 22 patients in the distraction group (7).

Clinical morbidities

Short-term complications occurred at a similar frequency between the two groups and are described in Table 5. They were successfully managed in all but one case of severe relapse in the distraction group, due to severe palatal scarring. The authors acknowledged that the small sample size may not reflect the true incidence of operative and post-operative complications (6).

Speech and velopharyngeal function

Outcomes related to speech and velopharyngeal function were assessed pre-operatively and at post1 (mean of 4 months post-op) and post2 (mean of 17 months post-op) (8). Nasoendoscopy examination was undertaken to assess velopharyngeal function, nasalance was assessed using a nasometer device, and perceptual speech assessment was undertaken to review resonance and nasal emissions. No statistically significant differences could be detected between the interventions with regard to the effect on any of the speech-related outcomes investigated.

Soft and hard tissue changes

The change in position of various cephalometric landmarks horizontally and vertically in relation to X and Y reference lines respectively were assessed from baseline to 6 months, 1 year and 2 years (9). It demonstrates that maxillary advancement achieved through either DO or CO can produce significant soft tissue improvement of the nose and upper lip. The current evidence however does demonstrate there are some aesthetic differences between the results achieved with the two techniques.

Following advancement of the maxilla in the distraction group the initial changes in the position of pronasale, labrale superius and subnasale were significantly greater than the control group. A significant difference in nasal projection was detected, with it initially being significantly more in the distraction group following both maxillary advancement and down grafting. But the study did not find a statistically significant difference in the nasolabial angle between the interventions. Maxillary advancement by DO also generates more consistent hard to soft tissue ratios than CO. For the majority of soft tissue parameters measured, the amount of change following maxillary advancement was greater in the distraction group.

Psychological adjustment

Psychological adjustment following the two interventions was assessed up to 2 years postoperatively using three self-reported questionnaires (10). Patients going through both

						No. of	Risk of
Paper	Outcome	Subdivision	Results in DO group	Results in CO group	Overall effect	participants	bias
Cheung	Clinical		1 patient relapsed to a class	1 patient relapsed to an edge	Short-term complications occurred at	29	High
GL al. (0)			2 participants developed	1 participant suffered sinusitis.	groups. There was no significant		
			mucosal infection around	1 individual experienced intra-	harm to any patient.		
			distractor.	operative bleeding.			
Cheung	Skeletal relapse	Short-term	Horizontal movement:	Horizontal movement	Further forward and downward	29	High
et al. (6)	of maxilla	skeletal	A point = $+3.7$ mm	A point $= -3.5$ mm	movement in the DO group.		
		relapse at	P point = $+2.4$ mm	P point= -1.8 mm	Backward and upward relapse in		
		1 year post-	Vertical movement:	Vertical movement:	the CO group. Authors did not		
		operatively	A point = $+3.3$ mm	A-point = -2.7 mm	assess if this was statistically		
			P point = $+1.8 \text{ mm}$	P point = -2.2 mm	significant.		
Chua	Skeletal relapse	Long-term	Horizontal movement:	Horizontal movement	Statistically significant difference in	47	High
et al. (7)	of maxilla	skeletal	A point = +2.27 mm (+32.3%)	A point = -2.53 mm (-36.9%)	relapse of A and P points between		
		relapse at	P point = +2.51 mm (+35.5%)	P point= -2.54 mm (-35.5%)	the two surgical intervention along		
		5 years post-	Vertical movement:	Vertical movement:	the horizontal plane.		
		operatively	A point = +1.09 mm (+83.2%)	A-point = $-0.4 \text{ mm} (-24.8\%)$	Relapse greater in the CO group		
			P point = +0.25 mm (+25.8%)	P point = -0.89 mm (-101.1%)	compared to the DO group 5 years		
					post-op.		
Chua	Speech	VP function at	Improved 3/10 participants	Improved 2/11 participants	RR improved VP status=1.650	21	High
et al. (8)		post2	Deteriorated 2/10 participants	Deteriorated 2/11 participants	(95% CI 0.343-7.939)		
					RR deteriorated VP status = 1.10		
					(95% CI 0.189-6.413)		
					No SS diff between interventions		
					in VP status.		
Chua	Speech	Hypernasality at	Improved 0/11 participants	Improved 0/11 participants	RR hypernasality improving = 1	22	High
et al. (8)		post2	Deteriorated 0/11 participants	Deteriorated 4/11 participants	(95% CI 0.022-46.408)		
					RR hypernasality deteriorating = RR		
					0.111 (95% CI 0.007–1.846).		
					No SS diff between interventions for		
					resonance.		

Table 5. Summary of findings for the publications included within the systematic review

Paper	Outcome	Subdivision	Results in DO group	Results in CO group	Overall effect	No. of participants	Risk of bias
Chua et al. (8)	Speech	Nasal emissions at post2	Improved 0/11 participants Deteriorated 1/11 participants	Improved 0/11 participants Deteriorated 1/11 participants	RR improved nasal emissions = 1 (95% Cl 0.022-46.408). RR deteriorated nasal emissions = 1 (95% Cl 0.071-14.053). No SS diff between groups for nasal emissions.	52	H
Chua et al. (8)	Speech	Nasalance at post2.	Mean change pre-op to post2 = -1.62%	Mean change pre-op to post2 = 8.6%	No SS difference between interventions for nasalance scores.	20	High
Chua et al. (10)	Psychological adjustment	SADS			No SS differences between the SADS scores for the two interventions.	30	High
Chua et al. (10)	Psychological adjustment	CFSEI			CFSEI-social score in DO group at 2-8 weeks and 3 months post-op	30	High
Chua et al. (10)	Psychological adjustment	SMLS			SWLS score at 1 and 2 years post-op are statistically significantly greater in DO group.	30	High
Chua and Cheung (9)	Soft and hard tissue changes	Change in lip thickness			No SS diff in the lip thickness between two groups following maxillary advancement and down grafting.	66	High
Chua and Cheung (9)	Soft and hard tissue changes	Soft tissue landmark change			Maxillary advancement: Maxillary advancement: SS diff between interventions following maxillary advancement: S1: dPN, dSn, dLS S2: dPn S3: dLS S3: dLS All changes greater in the DO group.	б. б.	Hgh
					No SS diff between interventions.		

Table 5. (continued)

Paper	Outcome	Subdivision	Results in DO group	Results in CO group	Overall effect	No. of participants	Hisk of bias
Chua and Cheung (9)	Soft and hard tissue changes	Change of nasolabial angle			No SS diff in change of nasolabial angle between the two groups following maxillary advancement	68	High
Chua and Cheung (9)	Soft and hard tissue changes	Change of nasal projection			Maxillary advancement: Maxillary advancement: SS diff in change of nasal projection at S1 and S2. Greater in DO group. Maxillary down grafting: SS diff in change of nasal projection	б Ю	H
Chua and Cheung (9)	Soft and hard tissue changes	Hard to soft tissue ratios	Maxillary advancement: Nearly all hard to soft tissue ratios at all follow-up time periods were significantly correlated. Maxillary down gratting: Significant correlations were: S2: UI: Ls S3: UI:Ls, UI:Stms	Maxillary advancement: The majority of significant hard to soft tissue ratios occurred during S1 and S2. The only ratio not significant at any time point was A point: Stms. Maxillary down grafting: None of the ratios were statistically significant.	at S1. Greater in D0 group. Following maxillary advancement the soft tissue changes achieved by distraction osteogenesis were generally greater than those achieved by orthognathic surgery. Following maxillary down grafting the hard and soft tissue ratios of both interventions were not well correlated.	е е	H

Table 5. (continued)

interventions experienced a reduction in social avoidance and distress, but this decrease was not statistically significant.

Social self-esteem improved following orthognathic surgery. In the distraction group during the early post-operative period social self-esteem fell by a statistically significant amount compared to the control intervention. At 6 months post-operatively social self-esteem then started to rise, reaching a level of self-esteem similar to that of the osteotomy patients.

The well-being of the individuals was measured with the satisfaction with life scale (SWLS). The osteotomy group considered themselves to be 'slightly satisfied' with life at every follow-up period. Pre-operatively the distraction group were 'slightly satisfied' and there was a gradual rise in SWLS scores from 3 months onwards. At 2 years post-operatively the SWLS shifted from 'slightly satisfied' to 'satisfied' with life in the DO group. Comparison of the SWLS scores for the two interventions found that life satisfaction was statistically significantly greater in the distraction group at 1 and 2 years post-operatively.

Discussion

In this systematic review five publications relating to a single randomized controlled trial were identified comparing the effectiveness of internal distraction osteogenesis to conventional orthognathic surgery for the correction of moderate maxillary hypoplasia in individuals with cleft lip and palate. The search process however illustrated that in this area the majority of the research still consists predominantly of case series and retrospective study designs.

Effect of intervention

Following surgical correction of maxillary hypoplasia the main aims of treatment are to achieve a well-balanced face and normal occlusion. The findings of the review suggest that both internal distraction osteogenesis and conventional osteotomy can produce significant soft tissue improvement of the lip and nose, although there are some aesthetic differences between the results achieved with the two techniques (9).

In addition there appears to be a difference between the two surgical interventions in relation to skeletal stability of the maxilla (6,7). Significantly more horizontal relapse of the maxilla occurred following orthognathic surgery compared to distraction osteogenesis. The backward relapse in the osteotomy group was most rapid up to 1 year post-operatively and then started to slow (7).

No difference could be detected between the different interventions with regard to the effect on speech and velopharyngeal status. In addition no correlation was detected between the amount of maxillary advancement and the likelihood of speech and velopharyngeal complications. The adaption of the velopharyngeal mechanism following surgical advancement is unpredictable and appears to be variable. A recommendation made by the trial authors is that all patients with cleft lip and palate being considered for maxillary advancement by either technique should be counselled prior to surgery about the unpredictable effects on speech and velopharyngeal function (8).

With respect to psychological adjustment, distraction osteogenesis in the early post-operative period lowers the social self-esteem and confidence of patients. In the long term however it results in better life satisfaction when compared to conventional orthognathic surgery. It is therefore suggested that patients receiving distraction osteogenesis are provided with psychological support to overcome the initial anxiety associated with this treatment modality (10).

Quality of evidence

All the publications featured within this review have a high risk of bias in at least one domain of the Cochrane Collaboration's risk of bias tool, resulting in a high overall risk of bias. The evidence included within this review is therefore of limited quality with significant biases associated with study quality.

Assessment of study quality was complicated by incomplete and contradictory reporting between the five publications. Deficiencies related to the quality of reporting are common (20), but this does not necessarily always reflect the conduct of the study (21).

Limitations of the review

This review is based upon the findings of what appears to be a single trial which treated a narrow range of participants with specific disease characteristics. This therefore affects the generalizability of the data to other groups. All the included papers focus on internal distraction, ignoring alternative distraction treatment protocols such as external distraction. In addition this review highlights the lack of high quality studies in this area. The difficulty faced with all developing techniques is that just because a technique is not supported by high quality evidence, it can only be concluded that there is insufficient evidence and not that the technique is ineffective. In addition the control of multiple variables necessary for such randomized controlled trials make such studies difficult to accomplish and therefore in some cases may never be conducted.

To help gather evidence in such areas one suggestion would be that all published clinical cases, series and observational studies should be reported in a standardized format established by experts, similar to the CONSORT guidelines for RCTs. Or alternatively cases should be registered in a prospective standardized registry, such as the EUROCRAN distraction study. This would hopefully make it easier to analyse the experience of clinicians and allow a consensus regarding a treatment modality to be made. This may allow an idealized treatment protocol to be formulated, which future randomized controlled trials could be based upon.

Despite significant efforts nine publications identified during the search process were inaccessible. This consisted of grey literature in the form of conference proceedings and academic theses (Fig. 1). It has been shown that published trials show a greater overall treatment effect than grey literature (22). Therefore failure to access this literature may have resulted in bias and skewed the results of the review process to favour the experimental intervention (5).

One of the inclusion criteria within this review was that the publication must be written in English, leading to possible reporting bias. In total, 52 of the papers identified during the search process were not written in English. Despite this the majority of these manuscripts had titles and abstracts translated into English. Review of these translations indicates that application of all the other inclusion criteria would have meant that 49 of the papers would have been excluded from the review because in addition to the main manuscript not being in English, there were also further deficiencies in the studies. There were three publications which were solely excluded because they were not written in the English language.

Authors are more likely to report outcomes in an international, English-speaking journal if the findings are positive (23). In contrast, alternative research (24) has found overall the exclusion of trials not reported in English does not significantly alter the results of meta-analyses.

Overall, the robustness of the findings of this review are constrained by the limited data available for synthesis. In addition the quality of the data is restricted by the high risk of bias of the included publications.

Implications for practice

In conclusion there is weak evidence that the effectiveness of distraction osteogenesis and conventional osteotomy for the treatment of cleft-related maxillary hypoplasia may differ for certain outcomes. The publications included within this review found that horizontal skeletal stability was improved following internal maxillary distraction. In addition whilst there may be an initial fall in self-confidence and self-esteem with this form of surgical correction, it improves with time. The evidence included within this review however must be interpreted with caution given the limited and relatively low quality and potential bias of the publications. Therefore currently there is insufficient evidence to make definite conclusions comparing the effectiveness of distraction osteogenesis to orthognathic surgery for the correction of maxillary hypoplasia

secondary to cleft lip and palate and no implications for practice can be made.

Implications for research

There is a need for further research in the long term in the form of prospective randomized controlled trials with adequate sample sizes and reduced bias, to allow conclusive recommendations to be made. These should compare the effectiveness of distraction osteogenesis against maxillary osteotomy and also compare different types of alveolar distraction such as external, internal and anterior maxillary distraction.

This review highlights the deficiencies in the standard of reporting of studies in this area. Any future trials should be designed, carried out and reported according to the Consolidated Standards of Reporting Trials (CONSORT) guidelines (25). This should ensure future trials are of a high quality with good validity.

Conclusion

There is a lack of sufficient evidence to conclude whether distraction osteogenesis is more

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or less effective than conventional orthognathic surgery for the treatment of cleft-related maxillary hypoplasia. The five publications included within this review, all reporting different outcome measures of a single trial, were at high risk of bias and no implications for practice can therefore be given.

Clinical relevance

Both maxillary osteotomy and distraction osteogenesis are accepted treatment modalities for cleft-related maxillary hypoplasia. This systematic review found weak evidence that the horizontal skeletal stability of the maxilla was improved following internal distraction compared to conventional osteotomy. Also whilst there may be an initial fall in self-confidence and self-esteem with distraction osteogenesis, it improves with time. The evidence included within this review however must be interpreted with caution given the limited and relatively low quality of the included publications. Therefore currently there is insufficient evidence to recommend the effectiveness of one technique over the other.

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