Comparison of adverse effects between lingual and labial orthodontic treatment

A systematic review

Hu Long^a; Yang Zhou^b; Ujjwal Pyakurel^b; Lina Liao^a; Fan Jian^a; Junjie Xue^a; Niansong Ye^a; Xin Yang^a; Yan Wang^c; Wenli Lai^d

ABSTRACT

Objective: To compare adverse effects between labial and lingual orthodontic treatments through a systematic review of the literature.

Materials and Methods: The protocol of this systematic review (CRD42012002455) was registered in the International Prospective Register of Systematic Reviews (PROSPERO). An electronic search was conducted in PubMed, Embase, Web of Science, CENTRAL, SIGLE, ProQuest Dissertations & Theses, and ClinicalTrial.gov for articles published between January 1980 and December 2012. Primary outcomes included pain and caries; secondary outcomes were eating difficulty, speech difficulty, oral hygiene, and treatment duration. Meta-analyses were conducted in Comprehensive Meta-Analysis version 2.2.064.

Results: Six studies were included, two randomized controlled trials and four clinical controlled trials; of these, four were medium quality and two were low quality in terms of the risk of bias. Five of the six outcomes were evaluated in the included studies, and treatment duration was not; pain, eating difficulty, speech difficulty were statistically pooled. Meta-analysis revealed that the pooled odds ratios were 1.20 (95% confidence interval [CI] = 0.30-4.87) for overall pain, 32.24 (95% CI = 14.13-73.55) for pain in tongue, 0.08 (95% CI = 0.04-0.18) for pain in cheek, 0.11 (95% CI = 0.03-0.42) for pain in lip, 3.59 (95% CI = 1.85-6.99) for eating difficulty, and 8.61 (95% CI = 3.55-20.89) for speech difficulty. Sensitivity analysis showed consistent results except for eating difficulty. No publication bias was detected.

Conclusions: The likelihood of overall pain was similar between the two modalities. Patients who underwent lingual orthodontic treatment were more likely to suffer from pain in the tongue and less likely to suffer from pain in the cheek and lip. Lingual orthodontic treatment increased the likelihood of speech difficulty. Eating difficulty, oral hygiene, caries, and treatment duration could not be compared in this systematic review. (*Angle Orthod.* 2013;83:1066–1073.)

KEY WORDS: Lingual orthodontics; Labial orthodontics; Meta-analysis; Systematic review; Adverse effect

INTRODUCTION

Since the advent of lingual orthodontic appliances in the 1970s,¹ recent years have witnessed a marked increase in the demand for lingual orthodontic appliances among orthodontic patients seeking esthetic improvement.² Several seminal studies indicated that lingual appliances can provide treatment outcomes comparable to those achieved with labial appliances.^{3,4} Lingual orthodontic appliances enjoy esthetic advantages

^a PhD Student, State Key Laboratory of Oral Diseases, Department of Orthodontics, West China Hospital of Stomatology, Sichuan University, Chengdu, China.

^b MS Student, State Key Laboratory of Oral Diseases, Department of Orthodontics, West China Hospital of Stomatology, Sichuan University, Chengdu, China.

^c Lecturer, State Key Laboratory of Oral Diseases, Department of Orthodontics, West China Hospital of Stomatology, Sichuan University, Chengdu, China.

^d Professor, State Key Laboratory of Oral Diseases, Department of Orthodontics, West China Hospital of Stomatology, Sichuan University, Chengdu, China.

Hu Long and Yang Zhou contributed equally to this work.

Corresponding author: Professor Wenli Lai, State Key Laboratory of Oral Diseases, Department of Orthodontics, West China Hospital of Stomatology, Sichuan University, No. 14, Section 3, Ren Min Nan Road, Chengdu 610041, China (e-mail: wenlilai@hotmail.com)

Accepted: March 2013. Submitted: January 2013.

Published Online: April 12, 2013

 $^{{\}scriptstyle \circledcirc}$ 2013 by The EH Angle Education and Research Foundation, Inc.

Table 1.	Search	Strategies	for	Each	Database ^a

Database	Search Strategy
PubMed	(orthodontics [mesh] OR orthodontic*) AND (labial OR labio OR "labial orthodontics" OR "labial appliance" OR "labial bracket" OR buccal OR bucco OR "buccal orthodontics" OR "buccal appliance" OR "buccal bracket") AND (lingual OR linguo OR "lingual orthodontics" OR "lingual appliance" OR "lingual bracket")
Embase	orthodontic* AND (labial OR labio OR "labial orthodontics" OR "labial appliance" OR "labial bracket" OR 'buccal/ exp OR buccal OR bucco OR "buccal orthodontics" OR "buccal appliance" OR "buccal bracket") AND ('lingual/ exp OR lingual OR linguo OR "lingual orthodontics" OR "lingual appliance" OR "lingual bracket")
Web of Science	orthodontic* AND (labial OR labio OR "labial orthodontics" OR "labial appliance" OR "labial bracket" OR buccal OR bucco OR "buccal orthodontics" OR "buccal appliance" OR "buccal bracket") AND (lingual OR linguo OR "lingual orthodontics" OR "lingual appliance" OR "lingual bracket")
CENTRAL	orthodontic* AND (labial OR labio OR "labial orthodontics" OR "labial appliance" OR "labial bracket" OR buccal OR bucco OR "buccal orthodontics" OR "buccal appliance" OR "buccal bracket") AND (lingual OR linguo OR "lingual orthodontics" OR "lingual appliance" OR "lingual bracket")
ProQuest Dissertations & Theses SIGLE ClinicalTrial.gov	diskw(orthodontic) AND [diskw(labial) OR diskw(buccal)] and diskw(lingual) orthodontic AND (labial OR buccal) AND lingual orthodontic AND (labial OR buccal) AND lingual

^a Limits: publication date between January 1980 and December 2012.

over conventional labial orthodontic appliances.⁵ Moreover, it has been claimed that lingual appliances bear a lower risk of caries.⁶ Nevertheless, strong concerns regarding tongue soreness and difficulty in speech have arisen regarding lingual orthodontic appliances.^{7–10} Specifically, it was recently reported by Khattab et al.¹¹ that more significant speech deteriorations were associated with lingual orthodontic treatment than labial appliances. However, to date, the reliability of this evidence has not been critically assessed. Therefore, a systematic review that critically evaluates the reliability of evidence is necessary for relevant dental practitioners. We conducted a systematic review to compare adverse effects between lingual and labial orthodontic treatment among orthodontic patients.

MATERIALS AND METHODS

Registration of Systematic Review

The protocol for this systematic review was registered in the International Prospective Register of Systematic Reviews (PROSPERO) (http://www.crd.york.ac.uk/ prospero/) (registration number CRD42012002455).

Inclusion Criteria

Participants had to be healthy adults or children who had a certain type of dental malocclusion and required orthodontic treatment. Participants with orofacial anomalies (eg, cleft lip and palate), dental pathologies (eg, cyst), and/or medical conditions (eg, osteoporosis) had to be excluded. Included studies must have examined lingual and labial orthodontic treatment; only those studies that compared the two interventions were included. Primary outcomes included pain and caries; secondary outcomes were eating difficulty, speech difficulty, oral hygiene, and treatment duration. Both randomized controlled trials (RCTs) and controlled clinical trials (CCTs) were eligible.

Search Methods

We searched the databases PubMed, Embase, Web of Science, CENTRAL, ProQuest Dissertations & Theses, and ClinicalTrial.gov. Moreover, SIGLE was searched for grey literature. Hand searching was not performed. The specific search strategies are shown in Table 1. The electronic search included all articles published between January 1980 and December 2012, with no language restrictions. Two review authors conducted the electronic searches independently, and any disagreements were solved by discussion or judged by a third reviewer.

Data Extraction and Analysis

Resultdata regarding study design, participant information, intervention type, follow-up periods, and outcomes were extracted and recorded independently and in duplicate by two review authors. Any disagreement was solved by discussion or judged by a third author.

Moreover, the risk of bias for all the included studies were assessed independently and in duplicate by two review authors according to the Cochrane Collaboration tool for assessing risk of bias.¹² Specifically, the main items included: (1) adequate sequence generation; (2) allocation concealment; (3) blinding; (4) management of incomplete outcome data; (5) absence of selective reporting; and (6) absence of other sources of bias. Studies with four or more items, with high risk of bias were excluded from analyses.

All the meta-analyses were performed in Comprehensive Meta-Analysis (version 2.2.064, Biostat, Englewood, NJ). For dichotomous data, odds ratios (ORs)

			Participa	antsª		A	llocationª			Bracket	System
Study	Study Design	No.	M/F	Age, y	LI	LA	Group 1	Group 2	Follow-up	LI	LA
Caniklioglu and Ozturk (2005) ⁸	ССТ	60	21/39	17.9	30	30	-	-	3 mo	7th Generation, Ormco	Roth
Van der Veen et al. (2010) ⁶	RCT	28	N/A	18	-	-	14	14	$18.1\pm5.5\text{mo}$	Incognito, Top Service	Orthos, Ormco
Wu et al. (2010) ⁹	CCT	60	22/38	20.98	30	30	-	-	3 mo	Incognito	Mini-Diamond, Ormco
Wu et al. (2011)10	CCT	60	22/38	20.98	30	30	-	-	3 mo	Incognito	Mini-Diamond, Ormco
Shalish et al. (2012)19	CCT	47	18/29	18–60	19	28	-	-	14 d	Incognito	Ormco
Khattab et al. (2012) ¹¹	RCT	34	13/21	21.3 ± 3.1	17	17	-	-	3 mo	Stealth, American Orthodontics, Sheboygan, WI	Roth, Ormco, Orange, CA, USA.

Table 2. General Information Provided in the Included Studies

^a M indicates male; F, female; N/A, not available; LI, lingual orthodontic treatment; LA, labial orthodontic treatment; group 1, LI in upper arch and LA in lower arch; and group 2, LA in upper arch and LI in lower arch.

(with 95% confidence intervals [CIs]) were used for statistical pooling; for continuous data, standardized mean differences were first converted to ORs through the formula of Chinn.^{13,14} With this method, dichotomous and continuous data could be pooled together by means of ORs in this systematic review. Heterogeneity across studies was assessed through the l² statistic, and an l² statistic greater than 50% was considered a sign of substantial heterogeneity. If substantial heterogeneity existed, a meta-regression or subgroup analysis would be employed to explore the potential heterogeneity.

The tests of Egger et al.¹⁵ and Begg and Mazumdar,¹⁶ along with the "trim and fill" method,^{17,18} were used to evaluate publication bias. Furthermore, sensitivity analysis was performed to evaluate the robustness of the pooled results from the meta-analysis. Cumulative meta-analysis was performed to determine the chronological changes in the pooled results from the year of first publication to the most recent publication.

RESULTS

Description of Studies

The agreement between the two independent reviewer authors with regard to article screening was

almost perfect (kappa = 0.922). Initially, we identified 718 articles from the database and excluded 708 as irrelevant. The remaining 10 articles were further assessed for eligibility, and six studies (two RCTs and four CCTs) were finally included in this review.^{6,8–11,19} The sample size ranged from 28 to 60 and the treatment durations ranged from 14 days to 18 months. One study⁶ included adolescents, while the other five included adults. Four articles^{6,8,9,11} were of medium quality and two^{10,19} were of low quality. The procedures of electronic searching are shown in Figure 1. The details of each study and the risks of bias are presented in Tables 2 and 3, respectively. All six included studies were prospective in nature.

Description of Outcomes

Of the six outcomes proposed for investigation, five (pain, eating difficulty, speech difficulty, oral hygiene, and caries) were evaluated, while one (treatment duration) was not evaluated in any of the included studies.

Description of Interventions

Brackets and archwires were located on the lingual surfaces of the teeth for lingual orthodontic treatment

Table 3. Risk of Bias of the Included Studies^a

Study	Item 1	Item 2	Item 3	Item 4	Item 5	Item 6	Score ^b	Quality
Caniklioglu and Ozturk (2005)8	Unclear	Unclear	High	Low	Low	Unclear	7	Medium
Van der Veen et al. (2010)6	Low	Unclear	High	High	High	Low	5	Medium
Wu et al. (2010) ⁹	High	High	High	Low	Low	Unclear	5	Medium
Wu et al. (2011) ¹⁰	High	High	High	Unclear	Low	Unclear	4	Low
Shalish et al. (2012)19	High	High	High	Unclear	Low	Unclear	4	Low
Khattab et al. (2012)11	Low	Low	High	Low	Low	High	8	Medium

^a Item definitions: Item 1 indicates adequate sequence generation; item 2, allocation concealment; item 3, blinding; item 4, management/ discussion of incomplete outcome data; item 5, absence of selective reporting; and item 6, absence of other apparent bias.

^b Scoring rules: Low indicates a score of 2; Unclear, a score of 1; and High, a score of 0. Quality was categorized as low (score 1–4), medium (score 5–8), or high (score 9–12).

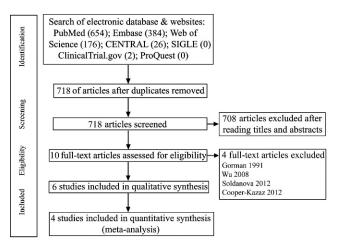


Figure 1. PRISMA flow diagram for studies retrieved through the search and selection processes. Gorman and Smith (1991)³ and Soldanova et al. (2012)²⁰ were excluded for nonextractable data, Wu et al. (2008)²¹ was excluded because its results were similar to those of Wu et al. (2010),⁹ and Cooper-Kazaz et al. (2012)²² was excluded because pain data had already been published in Shalish et al. (2012).¹⁹

and were located on the labial surfaces for labial orthodontic treatment.

Study Outcomes

Pain. Four studies^{8,9,11,19} investigated this outcome. Caniklioglu and Ozturk⁸ and Wu et al.⁹ specified the location of pain, ie, cheek, lip, and tongue, while Shalish et al.¹⁹ and Khattab et al.¹¹ did not. In addition, Canikioglu and Ozturk,8 Wu et al.,10 and Khattab et al.11 evaluated eating difficulty at 3 months, while Shalish et al.¹⁹ evaluated this factor after only 2 weeks. However, all four studies were included in the initial metaanalysis, and we then performed a sensitivity analysis that excluded Shalish et al.¹⁹ With respect to overall pain, the meta-analysis revealed that the pooled OR for overall pain was 1.20 (95% CI = 0.30-4.87)(lingual, n = 96; labial, n = 105) (Figure 2). As shown in Table 4, the sensitivity analysis that excluded Shalish et al.¹⁹ and the low-quality studies resulted in no significant changes in the pooled results, indicating the robustness of the original estimate of the metaanalysis. Moreover, two studies^{8,9} specified pain levels in specific locations, ie, tongue, cheek, and lip. As shown in Figure 2, the meta-analysis showed that the pooled ORs (lingual, n = 60, versus labial, n = 60) were 32.24 (95% CI = 14.13–73.55), 0.08 (95% CI = 0.04-0.18), and 0.11 (95% CI = 0.03-0.42) for pain in the tongue, cheek, and lip, respectively.

Eating difficulty. Four studies^{8,10,11,19} investigated this outcome. Canikioglu and Ozturk,⁸ Wu et al.,¹⁰ and Khattab et al.¹¹ evaluated eating difficulty at 3 months, while Shalish et al.¹⁹ evaluated this factor at 2 weeks.

All four studies were included in the original metaanalysis, and then we performed a sensitivity analysis by excluding Shalish et al.¹⁹ The pooled OR (lingual, n = 96, versus labial, n = 105) for eating difficulty was 3.59 (95% CI = 1.85–6.99) (Figure 3). As displayed in Table 4, the sensitivity analysis that excluded Shalish et al.¹⁹ revealed no significant changes. However, the sensitivity analysis that excluded low-quality studies did result in significant changes.

Speech difficulty. Three studies^{8,10,11} examined this outcome. The pooled OR for speech difficulty (lingual, n = 77, versus labial, n = 77) was 8.61 (95% Cl = 3.55–20.89) (Figure 4). The sensitivity analysis did not reveal any significant change (Table 4).

Oral hygiene. Only one study investigated this outcome. Caniklioglu and Ozturk⁸ revealed that the frequencies of oral hygiene problems within the first 3 months of treatment were similar between the two modalities (risk ratio, lingual versus labial: 1.40 [95% CI = 0.91-2.15]). Specifically, this study showed that food impaction was significantly more prevalent in lingual orthodontics (risk ratio 1.25 [95% CI = 1.03-1.50]), whereas the prevalence of bleeding gums and bad taste were similar between the two modalities (risk ratios: 0.73 [95% CI = 0.34-1.55] and 0.71 [95% CI = 0.26-2.00], respectively).

Caries. Only one study⁶ investigated this outcome. It revealed that the incidences of new white spot lesions were significantly lower in lingual orthodontics than in labial orthodontics (21 lesions/28 patients vs 4 lesions/28 patients; P = .004). Moreover, this study employed quantitative light-induced fluorescence for quantification and revealed that caries extent changed from $0.9\% \cdot \text{mm}^2 \pm 109.78\% \cdot \text{mm}^2$ to $5.7\% \cdot \text{mm}^2 \pm 2.82\% \cdot \text{mm}^2$ for lingual orthodontics but changed from $8.2\% \cdot \text{mm}^2 \pm 27.54\% \cdot \text{mm}^2$ to $58.4\% \cdot \text{mm}^2 \pm 122.95\% \cdot \text{mm}^2$ for labial orthodontics. The paired *t*-test revealed that the differences between lingual and labial orthodontics were statistically significant (P = .03).

Treatment duration. Unfortunately, none of the included studies evaluated this outcome.

Sensitivity Analysis

The results of sensitivity analyses are shown in Table 4. Because Shalish et al.¹⁹ evaluated outcomes at 2 weeks, while the other studies evaluated outcomes at 3 months, we excluded Shalish et al.¹⁹ from the meta-analysis to perform a sensitivity analysis and found no significant change. Khattab et al.¹¹ treated only upper arches, but all other studies included both arches in orthodontic treatment. Thus, a sensitivity analysis that excluded Khattab et al.¹¹ was performed and resulted in no significant changes. Exclusion of low-quality studies revealed no significant

Study name	Statistics for each study					Odds ratio	and 95% CI		
	Odds ratio	Lower limit		Z-Value	p-Value				
1. Overall pain									
Caniklioglu 2005	3.10	0.12	79.19	0.68	0.49		I		-1
Wu 2010	0.68	0.27	1.71	-0.82	0.41		_	-	
Shalish 2012	3.97	1.33	11.82	2.48	0.01				
Khattab 2012	0.12	0.01	2.48	-1.37	0.17	- K		_	
Total	1.20	0.30	4.87	0.26	0.80				
Random effect m	odel (I	² =65%)				0.01 Fav	0.1 1 vours Lingual	10 Favours Labi	100 al
2. Pain in tongue							-		
Caniklioglu 2005	24.75	5.86	104.54	4.37	0.00	1	1 1		-
Wu 2010	36.67	13.41	100.29	7.02	0.00				\rightarrow
Total	32.24	14.13;	73.55	8.25	0.00				
Fixed-effect mod	lel (I ² =	:0%)				0.01 Fai	0.1 1 vours Lingual	10 Favours Labi	100 al
3. Pain in cheek						14	ours Elliguar	Lavours Labi	
Caniklioglu 2005	0.09	0.02	0.32	-3.68	0.00	1 -	-#-		- T
Wu 2010	0.08	0.03	0.21	-4.89	0.00				
Total	0.08	0.04	0.18	-6.12	0.00		♣		
Fixed-effect mod	del (I ² =	:0%)				0.01	0.1 1	10	100
4. Pain in lip						Far	vours Lingual	Favours Labi	al
Caniklioglu 2005	0.23	0.07	0.76	-2.42	0.02		+=-		- I
Wu 2010	0.06	0.02	0.17	-5.28	0.00	<u> </u>	╼┼╴│		
Total	0.11	0.03	0.42	-3.24	0.00	.	\rightarrow		
Random effect n	nodel ([² =64%])			0.01	0.1 1	10	100
						Fay	ours Lingual	Favours Labi	al

Figure 2. Forest plot of pooled ORs regarding overall pain, pain in tongue, pain in cheek, and pain in lip for lingual versus labial orthodontic treatment.

changes, except for eating difficulty. Furthermore, changes in effect models (fixed-effect or random effect model) failed to reveal any significant change.

Meta-regression or Subgroup Analysis

Substantial heterogeneity was detected only for overall pain ($I^2 = 65\%$) and pain in lip ($I^2 = 64\%$) (Figure 2). Meta-regression was employed to explore potential heterogeneity. Although the different bracket systems used in the included studies may increase clinical heterogeneity, which would influence treatment

effects significantly (eg, torque), this factor may not influence the outcomes of the present analysis. Thus, we did not perform a meta-regression on it. However, because of the limited number of studies that investigated lip pain (n = 2), meta-regression was performed only for overall pain. The meta-regression revealed that follow-up durations and quality of studies were significantly associated with the pooled ORs (both P = .01). Because the follow-up periods were 2 weeks in Shalish et al.¹⁹ and 3 months in other three studies that investigated overall pain, and because

Table 4.	Results of	Sensitivity	Analysis	(ORs and	95% Cls)
----------	------------	-------------	----------	----------	----------

			Exclusion of		Changes in I	iges in Effect Models		
Item	Original Estimates ^a	Exclusion of Shalish et al. (2012) ¹⁹	Khattab et al. (2012) ¹¹	Exclusion of Low- Quality Studies ^b	Fixed	Random		
Overall pain	1.20 (0.30, 4.87)	0.66 (0.28, 1.55)	1.75 (0.43, 7.18)	0.66 (0.28, 1.55)	1.30 (0.67, 2.54)	1.20 (0.30, 4.87)		
Pain in tongue	32.24 (14.13, 73.55)	-	-	-	32.24 (14.13, 73.55)	32.24 (14.13, 73.55)		
Pain in cheek	0.08 (0.04, 0.18)	-	-	-	0.08 (0.04, 0.18)	0.08 (0.04, 0.18)		
Pain in lip	0.11 (0.03, 0.42)	-	-	-	0.11 (0.05, 0.23)	0.11 (0.03, 0.42)		
Eating difficulty	3.59 (1.85, 6.99)	2.63 (1.15, 6.01)	3.38 (1.71, 6.69)	5.21 (0.83, 32.75)°	3.59 (1.85, 6.99)	3.59 (1.85, 6.99)		
Speech difficulty	8.61 (3.55, 20.89)	-	8.14 (3.21, 20.60)	23.87 (3.00, 190.16)	8.61 (3.55, 20.89)	8.61 (3.55, 20.89)		

^a For original estimates, a fixed-effect model was adopted for pain in tongue, pain in cheek, eating difficulty, and speech difficulty; a randomeffect model was adopted for general pain and pain in lip.

^b Low-quality studies were Wu et al.¹⁰ and Shalish et al.¹⁹

° Significant change from the original estimate.

Study name		Statis	tics for e	ach study			Odds ratio and 95% CI				
	Odds ratio	Lower limit	Upper limit	Z-Value	p-Value						
Caniklioglu 2005	3.22	0.32	32.87	0.99	0.32	1	- I -	+		· 1	
Wu 2011	2.21	0.87	5.58	1.67	0.09			H	-		
Shalish 2012	6.37	2.08	19.52	3.24	0.00						
Khattab 2012	11.67	0.58	235.94	1.60	0.11			-	-		
Total	3.59	1.85	6.99	3.76	0.00				\bullet		
Fixed-effect mod	lel (I ² =()%)				0.01	0.1	1	10	100	
						Fav	ours Ling	ıal	Favours Lab	ial	

Figure 3. Forest plot of pooled OR regarding eating difficulty for lingual versus labial orthodontic treatment.

Shalish et al.¹⁹ was of low quality while the other three were of medium quality, we excluded Shalish et al.¹⁹ from the meta-analysis and found no significant heterogeneity ($l^2 = 5\%$). Thus, we suggest that the risk of bias did not influence the validity of the pooled results. However, as mentioned earlier, since the sensitivity analysis that excluded Shalish et al.¹⁹ resulted in no significant changes in the pooled results, we decided not to exclude Shalish et al.¹⁹ from the meta-analysis.

Cumulative Meta-analysis

As displayed in Figure 5, overall pain was found to be similar between lingual and labial orthodontic treatment in studies published since 2005. Eating difficulty was revealed to be significantly different between these groups in studies published since 2012, and speech difficulty was found to be significantly different between the groups in studies published since 2005.

Assessment of Publication Bias

Because of the limited number of studies that analyzed pain in the tongue, cheek, and lip, assessment of publication bias was possible only for overall pain, eating difficulty, and speech difficulty. As shown in Table 5, none of the three tests detected any evidence of publication bias.

DISCUSSION

In this systematic review, the six included studies evaluated five outcomes (pain, eating difficulty, speech difficulty, oral hygiene, and caries) in lingual and labial orthodontic treatment. Four studies were included in the meta-analysis of three outcomes (pain, eating difficulty, and speech difficulty). Sensitivity analysis showed consistent results in the meta-analysis, except for eating difficulty. Moreover, no evidence of publication bias was noted. Therefore, in general, the pooled results in the meta-analysis were robust.

The pooled OR for overall pain was 1.20 (95% CI = 0.30-4.87), indicating that the likelihood of overall pain was similar between lingual and labial orthodontic treatment. Although substantial heterogeneity existed for overall pain ($I^2 = 65\%$) and the meta-regression revealed that different follow-up durations and quality of studies could explain the heterogeneity (P = .01; $I^2 = 5\%$ after excluding Shalish et al.¹⁹ because of the short follow-up period and low quality), the sensitivity analysis that excluded Shalish et al.¹⁹ failed to reveal significant changes. Therefore, we decided not to exclude Shalish et al.¹⁹ in the meta-analysis for this factor. The meta-analysis showed that the pooled OR for pain in the tongue was 32.24 (95% CI = 14.13-73.55), indicating that patients receiving lingual orthodontic treatment would be more likely to suffer from pain in tongue than those receiving labial orthodontic treatment.

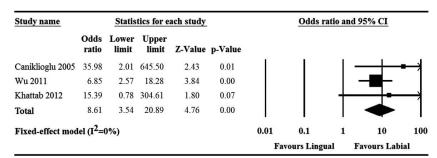


Figure 4. Forest plot of pooled OR regarding speech difficulty for lingual versus labial orthodontic treatment.

1071

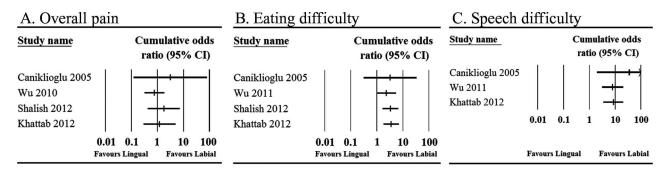


Figure 5. Cumulative meta-analysis of overall pain, eating difficulty, and speech difficulty.

Moreover, the pooled ORs were 0.08 (95% CI = 0.04-0.18) and 0.11 (95% CI = 0.03-0.42) for pain in the cheek and lip, respectively, indicating that patients would be less likely to suffer from pain in the cheek and lip when receiving lingual orthodontic treatment. For the meta-analysis of pain in the lip, substantial heterogeneity was detected ($I^2 = 64\%$). Ironically, no heterogeneity was detected in the meta-analyses of pain in the tongue ($I^2 = 0\%$) and pain in cheek ($I^2 =$ 0%), which included the same two studies (Caniklioglu and Ozturk⁸ and Wu et al.⁹). Therefore, we suggest that the heterogeneity between the two studies was random. However, because both individual studies revealed consistently similar results, which were also in line with the pooled results, we suggest that the pooled OR for pain in the lip is still reliable, regardless of the detected heterogeneity.

The pooled OR for eating difficulty was 3.59 (95% Cl = 1.85-6.99), indicating that patients undergoing lingual orthodontic treatment would be more likely to suffer from eating difficulty. Although no publication bias was noted, the sensitivity analysis that excluded low-quality studies resulted in a significant change (5.21, 95% Cl = 0.83-32.75), which prevented us from drawing a conclusion regarding differences in eating difficulty between the two modalities. Thus, we could not compare eating difficulty in this systematic review.

The pooled OR for speech difficulty was 8.61 (95% CI = 3.55-20.89), which was robust, as evidenced by the absence of significant changes in sensitivity analyses and absence of publication bias. Thus, we

suggest that speech difficulty would be more likely to occur during lingual orthodontic treatment.

In this systematic review, oral hygiene was evaluated in only one study (Caniklioglu and Ozturk⁸). This study revealed that the prevalence of oral hygiene problems was similar within the first 3 months between the two modalities (risk ratio [lingual versus labial] = 1.40 [95% CI = 0.91-2.15]). Although this study differentiated oral hygiene into food impaction, bleeding gums, and bad taste, it did not take the baseline data into consideration. Thus, we cannot compare oral hygiene between the two modalities in this systematic review.

In this systematic review, only one study⁶ compared caries between two modalities. This study counted the number of new white spot lesions through quantitative light-induced fluorescence. However, this finding considered the number but not the extent of new white spot lesions. Moreover, the statistical analysis was incorrect, since the paired *t*-test was used for data that obviously did not have a normal distribution (eg, $0.9\% \cdot mm^2 \pm 109.78\% \cdot mm^2$). Therefore, we could not compare caries between the two modalities in this systematic review.

The limitations of this systematic review include a lack of high-quality studies, small sample sizes, a limited follow-up period, flaws in statistical analysis in some of the included studies, and insufficient evidence for eating difficulty, oral hygiene, caries, and treatment duration. Specifically, it was reported that these adverse effects decreased gradually with time until removal of

Table 5.	Assessment of Publication Bias
----------	--------------------------------

	Test of Egger et al. ¹⁵	Test of Begg and		Trim and Fill Method ^a	
Item	(P Value)	Mazumdar ¹⁶ (P Value)	Original	Adjusted	Filled Studies
Overall pain	0.86	0.73	1.20 (0.30, 4.87)	1.20 (0.30, 4.87)	0
Eating difficulty	0.52	0.73	3.59 (1.85, 6.99)	3.38 (1.76, 6.48)	1
Speech difficulty	0.25	1.00	8.61 (3.54, 20.89)	6.85 (3.03, 15.49)	2

^a The "trim and fill" method utilizes an imputation of missing studies and fill these imputed missing studies into the funnel plot to make it symmetric. It adjusts the combined results by including these imputed studies. Thus, publication bias would be indicated if an adjusted estimate differed significantly from its original estimate and vice versa. In this present systematic review, since all of the adjusted estimates were similar to their original ones, the trim and fill method indicated no significant publication bias.

the brackets.⁷ Since the majority of the included studies followed patients for 3 months, the results of this systematic review should be interpreted with caution specifically as short-term effects. Therefore, more highquality studies, preferably RCTs, with larger sample sizes and longer follow-up periods are required.

CONCLUSION

- The likelihood of short-term overall pain was similar for labial and lingual orthodontic treatment.
- Patients receiving lingual orthodontic treatment were more likely to suffer from pain in the tongue but less likely to suffer from pain in the cheek and lip than those undergoing labial orthodontic treatment.
- Lingual orthodontic treatment bore a greater likelihood of speech difficulty.
- We could not compare eating difficulty, oral hygiene, caries, and treatment duration in this systematic review. Therefore, more high-quality studies, preferably RCTs, with larger sample sizes and longer follow-up periods are needed.

ACKNOWLEDGMENT

This work was supported by the National Natural Science Foundation of China (grants 81070858 and 81100778).

REFERENCES

- Fujita K. New orthodontic treatment with lingual bracket mushroom arch wire appliance. Am J Orthod. 1979;76: 657–675.
- Fritz U, Diedrich P, Wiechmann D. Lingual technique— Patients' characteristics, motivation and acceptance. Interpretation of a retrospective survey. *J Orofac Orthop.* 2002; 63:227–233.
- 3. Gorman JC, Smith RJ. Comparison of treatment effects with labial and lingual fixed appliances. *Am J Orthod Dentofacial Orthop.* 1991;99:202–209.
- 4. Gorman JC. Treatment of adults with lingual orthodontic appliances. *Dent Clin North Am.* 1988;32:589–620.
- 5. Fujita K. Development of lingual-bracket technique. (Esthetic and hygienic approach to orthodontic treatment) (Part 2) Manufacture and treatment [author's translation]. *Shika Rikogaku Zasshi.* 1978;19:87–94.
- van der Veen MH, Attin R, Schwestka-Polly R, Wiechmann D. Caries outcomes after orthodontic treatment with fixed appliances: do lingual brackets make a difference? *Eur J Oral Sci.* 2010;118:298–303.
- Miyawaki S, Yasuhara M, Koh Y. Discomfort caused by bonded lingual orthodontic appliances in adult patients as examined by retrospective questionnaire. *Am J Orthod Dentofacial Orthop.* 1999;115:83–88.

- Caniklioglu C, Ozturk Y. Patient discomfort: a comparison between lingual and labial fixed appliances. *Angle Orthod.* 2005;75:86–91.
- 9. Wu AK, McGrath C, Wong RW, Wiechmann D, Rabie AB. A comparison of pain experienced by patients treated with labial and lingual orthodontic appliances. *Eur J Orthod.* 2010;32:403–407.
- Wu A, McGrath C, Wong RW, Wiechmann D, Rabie AB. Comparison of oral impacts experienced by patients treated with labial or customized lingual fixed orthodontic appliances. *Am J Orthod Dentofacial Orthop.* 2011;139:784–790.
- Khattab TZ, Farah H, Al-Sabbagh R, Hajeer MY, Haj-Hamed Y. Speech performance and oral impairments with lingual and labial orthodontic appliances in the first stage of fixed treatment. *Angle Orthod.* 2012 Oct 18. [Epub ahead of print]
- Higgin JPT, Altman DG. Chapter 8: Assessing risk of bias in included studies. In: Higgin JPT, Green S, eds, *Cochrane Handbook for Systematic Reviews of Interventions 5.1.0* [updated March 2011]. The Cochrane Collaboration, 2011. Available at: handbook.cochrane.org. Accessed 14 March 2013.
- Chinn S. A simple method for converting an odds ratio to effect size for use in meta-analysis. *Stat Med.* 2000;19: 3127–3131.
- Deeks JJ, Higgin JPT, Altman DG. Chapter 9: Analysing data and undertaking meta-analyses. In: Higgin JPT, Green S, eds, *Cochrane Handbook for Systematic Reviews of Interventions 5.1.0* [updated March 2011]. The Cochrane Collaboration, 2011. Available at: handbook.cochrane.org. Accessed 14 March 2013.
- Egger M, Davey Smith G, Schneider M, Minder C. Bias in meta-analysis detected by a simple, graphical test. *BMJ*. 1997;315:629–634.
- Begg CB, Mazumdar M. Operating characteristics of a rank correlation test for publication bias. *Biometrics*. 1994;50: 1088–1101.
- Duval S, Tweedie R. Trim and fill: a simple funnel-plot-based method of testing and adjusting for publication bias in metaanalysis. *Biometrics*. 2000;56:455–463.
- Duval S, Tweedie R. A nonparametric "trim and fill" method of accounting for publication bias in meta-analysis. *J Am Stat Assoc.* 2000;95:89–98.
- Shalish M, Cooper-Kazaz R, Ivgi I, et al. Adult patients' adjustability to orthodontic appliances. Part I: a comparison between labial, lingual, and Invisalign. *Eur J Orthod*. 2012; 34:724–730.
- Soldanova M, Leseticky O, Komarkova L, Dostalova T, Smutny V, Spidlen M. Effectiveness of treatment of adult patients with the straightwire technique and the lingual twodimensional appliance. *Eur J Orthod.* 2012;34:674–680.
- 21. Wu AK, McGrath CP, Wong RW, Rabie AB, Wiechmann D. A comparison of pain experienced by patients treated with labial and lingual orthodontic appliances. *Ann R Australas Coll Dent Surg.* 2008;19:176–178.
- 22. Cooper-Kazaz R, Ivgi I, Canetti L, et al. The impact of personality on adult patients' adjustability to orthodontic appliances. *Angle Orthod*. 2013;83:76–82.