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The effects of oral-motor exercises on swallowing in children: an evidence-based systematic review

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ABSTRACT

Aim The aim of this unregistered evidence-based systematic review was to determine the state and quality of evidence on the effects of oral motor exercises (OME) on swallowing physiology, pulmonary health, functional swallowing outcomes, and drooling management in children with swallowing disorders.

Method A systematic search of 20 electronic databases was completed to identify relevant peer-reviewed literature published in English between 1960 and 2007. Experimental or quasi-experimental design studies examining OME as a treatment for children with swallowing disorders were appraised for methodological quality by two assessors and reviewed by a third.

Results Sixteen studies of varying methodological quality were included. No study examining the effects of OME on pulmonary health in children was identified. The included studies incorporated a wide variety of OME, and mixed findings were noted across all of the outcomes targeted in this review.

Interpretation Based on the results of this evidence-based systematic review, there is insufficient evidence to determine the effects of OME on children with oral sensorimotor deficits and swallowing problems. Well-designed studies are needed to provide clinicians with evidence that can be incorporated into the preferences of the client and the clinicians' knowledge of anatomy, physiology, and neurodevelopment in the management of this group of children.

List of Abbreviations

EBSR	Evidence-based systematic review
FFA	Functional Feeding Assessment
ISMAR	Innsbruck Sensorimotor Activator and Regulator
OME	Oral motor exercises

Swallowing disorders in children vary widely in terms of clinical presentation, etiology, severity, complexity, and impact on daily life.[1] These disorders range from transient and developmental to multidimensional and chronic or progressive.[2,3] For example, swallowing disorders are common in children with a variety of etiologies that include, but are not limited to, cerebral palsy (CP), genetic syndromes such as Down syndrome, and craniofacial anomalies. These swallowing disorders may involve multiple aspects of the feeding process (e.g. gathering food to the mouth, preparation, etc.), the swallowing process (e.g. bolus formation, oral phase abnormalities, impaired pharyngeal phase function, and reduced upper esophageal sphincter function), as well as difficulty with drooling or managing secretions.[4] Drooling (sialorrhea) most often occurs because of infrequent swallowing of saliva (secretions), and less frequently because of excess saliva production.[5,6] Moreover, these disorders may result in several health-related complications such as inadequate nutritional status and growth, reactive airway disease, and aspiration pneumonia.[7] Other children present with more mild developmental swallowing problems or oral-motor inefficiencies that may not have a negative impact on nutrition or overall health status. Although often considered less severe than other swallowing disorders, these oral motor disorders have been associated with dental malocclusion and mouth open resting posture.[8] Regardless of the etiology or severity, disruptions in the feeding and swallowing process may result in an increased burden to the caregiver, social restriction, and diminished quality of life.[9]

Because of the variability in ages and the types and severity of the feeding and swallowing problems, children with these disorders are seen in many settings (e.g. early intervention, preschool, and school-based) and may require different treatment approaches. Given these variables, speech-language pathologists will need the appropriate knowledge and skills to manage swallowing and feeding disorders across this diverse population.[10] Clinicians working with children who have feeding and swallowing problems frequently incorporate oral-motor exercises (OME) into their treatment plans.[11] There are three main categories of OME generally used in clinical practice: active exercises, passive exercises, and sensory applications.[12]

Active exercises include, but are not limited to, active range of motion, stretching, and strength training. These exercises are used to increase strength, endurance, and power through the recruitment of additional motor units as muscle fibers are enlarged.[13] Various forms of stretching affect muscle tone by manipulating the

muscle spindles either to inhibit or elicit a stretch reflex. By inhibiting this reflex through slow stretching, muscle tone may be reduced. By inducing a stretch reflex through quick stretch, tone is increased.

Passive exercises may include massage, stroking, stimulation, tapping, vibration, and passive range of motion exercises in which the movement is provided with the assistance of or entirely through the clinician or caregiver with little action from the individual receiving treatment. These procedures are applied to provide sensory input, improve circulation, and preserve or enhance joint flexibility. It has been theorized that some of these techniques normalize feeding patterns by reducing abnormal oral reflexes, facilitating normal muscle tone, or desensitizing the oral region.[14]

Sensory applications comprise the application of heat, cold, electrical stimulation, high-frequency vibration, or other agents to muscle tissues. Some (e.g. cold) may be used to enhance sensory awareness to initiate a swallow response. Others (e.g. electrical stimulation) are used to strengthen the swallowing musculature.

Although these techniques are widely used by clinicians, controversy exists about the theoretical soundness and effectiveness of these interventions for individuals with swallowing disorders.[12] Much of the debate has centered on the principle of training specificity, which argues that exercises that do not mirror the targeted function (swallowing) will not be effective in changing that target function. Therefore, OMEs that address underlying impairments (e.g. strength) but do not parallel the act of swallowing may not be effective in improving swallowing skills. Another factor contributing to this debate is the lack of normative data or objective and standardized measures to assess limitations targeted by OME, such as strength, endurance, and sensation, particularly in young children. Primary deficits in oral motor function involve weakness and incoordination. These deficits are typically inferred by clinical observation, not with objective measures. Thus, it is not possible to be objective in the perceptions of weakness and subsequent changes with varied OME. Moreover, to date, there are no widely accepted normative data in infants and children to define the necessary strength required to form a bolus and produce a swallow.

Some forms of OME have been examined. However, many of the findings are from a few small studies or produced mixed results.[13,15,16] In addition, these studies focused primarily on adult populations. Therefore, the effects of OME on the swallowing skills of children are unclear.

An important aspect of clinical decision making is the selection of appropriate intervention based on the three core principles of evidence-based practice: the preferences of the family and child, the clinician's expertise, and a consideration of the current best evidence.[17] Evidence-based clinical decision making is always important and it can identify areas where controversy exists, as in the case of OME. Practicing clinicians often report that they have insufficient time and resources to

search for and analyse the scientific literature to make evidence-based clinical decisions.[18] To assist clinicians, the American Speech-Language-Hearing Association's National Center for Evidence-based Practice began conducting evidence-based systematic reviews (EBSRs). These reviews use comprehensive and replicable methods to identify, evaluate, and synthesize the state of the evidence for a particular screening procedure, diagnostic tool, or intervention. The EBSR can assist multidisciplinary teams and purchasers by providing an understanding of the evidence in order to provide effective services. It can also highlight gaps in the current research and provide a focus for future research on a given topic. Given the extensive use of OMEs and the controversy surrounding them, the National Center for Evidence-based Practice and an expert panel initiated a series of systematic reviews on OME. The panel defined OME treatments as activities involving sensory stimulation to or actions of the lips, jaw, tongue, soft palate, larynx, and respiratory muscles that are intended to influence the physiological underpinnings of the oropharyngeal mechanism and thus improve its functions. This broad definition was developed to incorporate the three main categories of OME described above.

This series of reviews examined the use of OME across various populations (i.e. preterm infants, children, and adults) and multiple aspects of treatment by speech-language pathologists (i.e. speech and swallowing). The aim of this review was to determine the impact of OME on swallowing in children (other than preterm infants). It focused on four clinical questions: (1) What is the effect of OME on swallowing physiology (e.g. pressures, efficiency, aspiration, timing) in children? (2) What is the effect of OME on pulmonary health (i.e. aspiration pneumonia) in children? (3) What is the effect of OME on functional swallowing outcomes (e.g. oral feeding, volume intake, weight gain, growth) in children? (4) What is the effect of OME on drooling management in children?

A set of EBSRs (using a similar methodology) examining the impact of electrical stimulation on swallowing and OME treatments on speech are addressed in separate publications.[19,20]

METHOD

We conducted a single, systematic search of the literature for this unregistered review series between December 2006 and September 2007. Studies were initially considered for the review if they were published in a peer-reviewed journal from 1960 to 2007, were published in English (owing to limited translations resources), and contained original data addressing one or more of the clinical questions included in this series of EBSRs. Further inclusion criteria pertaining specifically to this review were studies incorporating an experimental or quasi-experimental design (including multiple baseline single-subject design investigations), studies conducted on children with swallowing disorders, and studies investigating the effects of OME as a treatment and not just a condition in which swallowing skills were examined. These study designs were included because they are generally

considered to demonstrate evidence of the causal effects of an intervention for a specific outcome. Studies focusing on preterm infants or those examining the use of neuromuscular electrical stimulation were excluded as these were the subjects of other reviews within this series. Studies that included surgical, medical, or pharmacological treatment were excluded. Studies that incorporated additional interventions paired with oral motor treatment, or used liquid or food as part of the oral motor intervention, were eliminated from consideration. These treatments were excluded because it is not possible to determine the impact of a specific intervention, in this instance OME, unless it is examined individually or controlled within a research design.

A set of expanded keywords was developed that related to oral motor exercises, swallowing, and speech. Table SI (published online only), provides a full list of expanded keywords and the detailed search strategy. The full author panel generated the core set of keywords. This initial set of keywords was intentionally broad to capture the span of interventions and outcomes addressed in the clinical questions targeted in this series of reviews. These keywords were then expanded based on the medical subject headings from the National Library of Medicine or the controlled vocabulary specific to each of the searched databases. Relevant abstracts and articles were also examined to identify additional search terms, keywords, and expanded medical subject headings.

Twenty electronic databases (Appendix SI, published online only), all American Speech-Language-Hearing Association journals, and Google Scholar were searched. Additionally, a hand search of references from all relevant articles was also completed to identify other applicable citations. Forward citation tracking of relevant articles was used to identify additional studies that cited those articles.

A total of 899 citations were initially identified for review (Fig. 1). Two authors, blind to one another's results, reviewed each abstract and initially identified 346 abstracts as meeting the inclusion criteria with 91% agreement. Of those preliminarily accepted, 250 were subsequently excluded because they did not directly address one or more of the larger set of clinical questions or report original data. The remaining 96 studies were identified for inclusion in this series of EBSRs. Of these, 16 studies addressed one or more of the four clinical questions related to the effects of OMEs on swallowing in pediatric populations and were identified for final inclusion in this review. A list of the excluded studies and the reason for their exclusion is available on request.

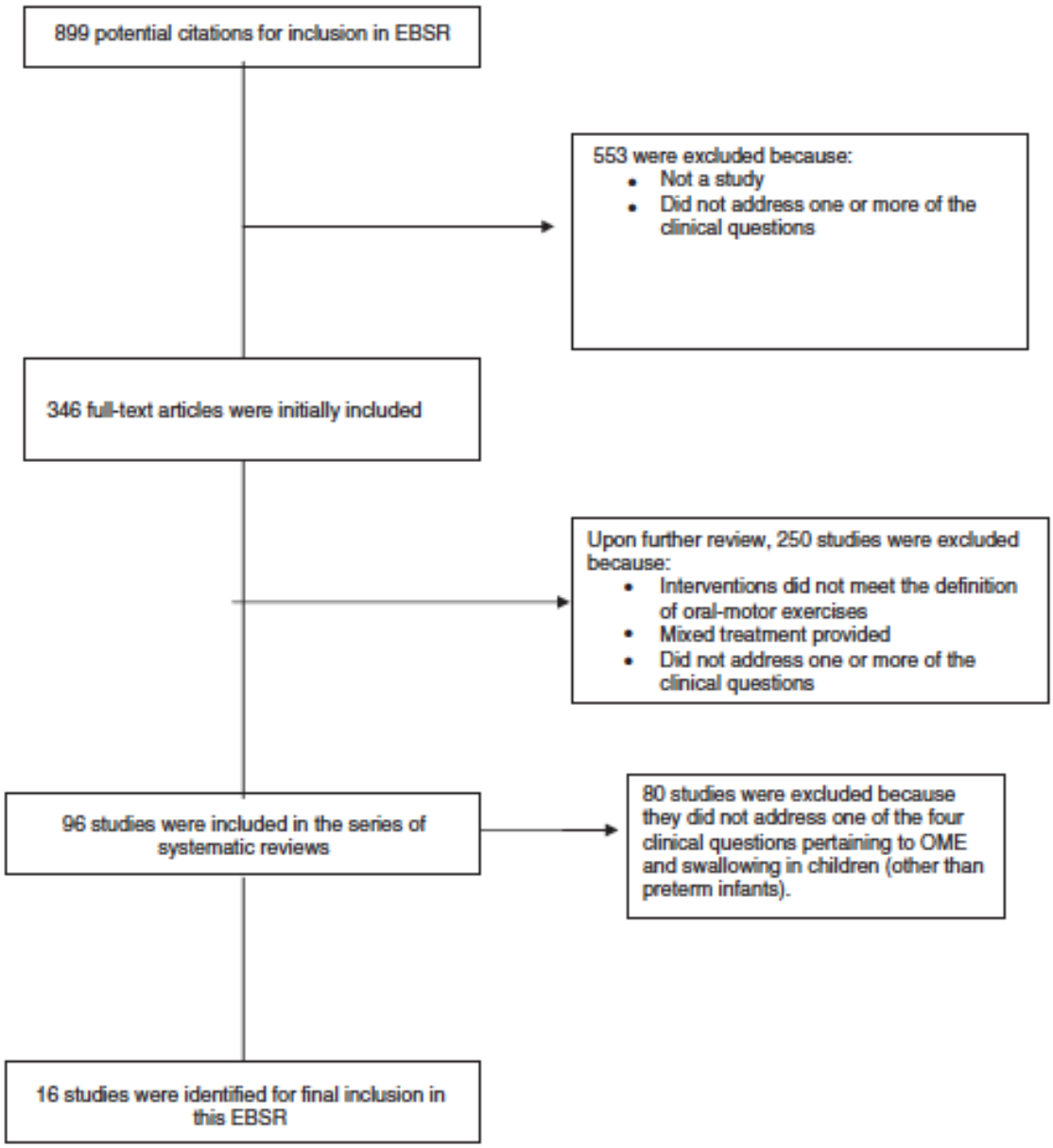

Figure 1: Process for identification of included studies. EBSR, evidence-based systematic review.

Included studies were then assessed for methodological quality based on the American Speech-Language-Hearing Association's Levels of Evidence Scheme.[21] This structured system was used to appraise each study across eight domains, which include the following: study design, assessor blinding, sampling/allocation, participant comparability/description, outcomes, significance, precision, and intention to treat (when applicable). These domains were selected to identify areas of possible bias or methodological characteristics that might influence estimates of treatment effects. Formal assessment of publication bias was not performed.

The two initial reviewers, still blind to one another's results, assessed each study for methodological quality (with 87% agreement) across the eight domains and determined a study quality marker score based on the number of indicators that met the highest level of quality in each area. A study received a point for each marker meeting the highest level of quality, as detailed in Table I. For studies incorporating controlled trials, all eight quality indicators were relevant, leading to a maximum quality score of 8. For all other study designs, where an intention-to-treat analysis was not applicable, the highest quality score was 7. A study was evaluated for each clinical question that it addressed. This was necessary because the targeted clinical questions and some of the appraisal domains were outcome-specific (e.g. significance, precision, etc.). Therefore, a study's quality marker score could vary depending upon which clinical question or outcome it targeted. For example, if a study reported a swallowing physiology outcome and a functional swallowing outcome but only reported the statistical significance of the physiology outcome, then the study would earn that appraisal point for clinical question 1 (the effect of OME on swallowing physiology in children) but not for clinical question 3 (the effect of OME on functional swallowing outcomes in children). Each critical appraisal was then reviewed by at least one member of the evidence panel who also completed the data extraction for the study. Agreement between the two initial reviewers and panel reviewers was greater than 98%. Discrepancies in ratings among authors were resolved by consensus.

Table I: Quality indicators

Indicator	Quality marker
Study design	Controlled trial[a]
	Cohort study
	Retrospective case–control or single-subject design
	Case series
	Case study
Blinding	Assessors blinded[a]
	Assessors not blinded or not stated
Sampling/ allocation	Random sample adequately described[a]
	Random sample inadequately described
	Convenience sample adequately described
	Convenience sample inadequately described or hand-picked sample or not stated
Group/ participant comparability	Groups/participants comparable at baseline on important factors (between-subject design) or participant(s) adequately described (within-subject design)[a]
	Groups/participants not comparable at baseline or comparability not reported or participant(s) not adequately described
Outcomes	At least one primary outcome measure is valid and reliable[a]
	Validity unknown, but appears reasonable; measure is reliable
	Invalid and/or unreliable
Significance	*p* value reported or calculable[a]
	p value neither reported nor calculable.
Precision	Effect size and confidence interval reported or calculable[a]
	Effect size or confidence interval, but not both, reported or calculable
	Neither effect size nor confidence interval reported or calculable.
Intention to treat (controlled trials only)	Analysed by intention to treat[a]
	Not analysed by intention to treat or not stated

[a]Indicates highest level of quality.

Effect sizes and corresponding confidence intervals were reported or calculated for outcome measures whenever possible. For group studies, Cohen's d was calculated from group means and standard deviations or estimated from results of analyses of variance or t-tests. The magnitude of effect sizes was determined using Cohen's benchmarks for small, medium, and large as 0.2, 0.5, and 0.8 respectively.[22]

A methodology proposed by Busk and Serlin[23] and described by Beeson and Robey[24] was used to calculate weighted effect-size estimates for multiple-baseline, single-subject design investigations. However, none of the included single-subject design studies provided sufficient data to perform these calculations.

RESULTS

Sixteen studies investigated the effects of OME on swallowing in children.[14,25–39] Eight studies examined the effects of OME on swallowing physiology (question 1), six studies examined functional swallowing outcomes (question 3), and five studies explored the effects of OME on drooling (question 4). This total exceeds 16 because several studies addressed multiple clinical questions. No study was found that examined the effect of OME on pulmonary health in children (question 2). The studies examined a total of 250 participants and included children with orofacial dysfunction and a tongue thrust pattern during swallowing (n=135), CP (n=86), Down syndrome (n=20), and multiple disabilities (n=9).

Clinical question 1: what is the effect of OME on swallowing physiology outcomes in children?

Table II summarizes the participant and intervention characteristics of the eight studies reporting data related to OME and swallowing physiology in children.[26,28,29,31–33,36,39] Of these, three examined the effects of an intra-oral stimulating plate, three investigated the impact of tongue thrust treatment or lip and tongue exercises, and two evaluated the use of oral, perioral, and facial stimulation.

Table II: Question 1: Swallowing physiology outcomes summary

Reference	*n*	Age in y:mo (range and/or mean)	Sex	Medical and/or speech-language pathology diagnosis	Intervention	Treatment schedule and amount	Outcome measure(s) and findings		Effect size (95% confidence interval)	Quality marker score
Christensen & Hanson[26]	10	5:8–6:9 (6:2)	6 males, 4 females	Severe anterior tongue thrust, acoustically severe frontal lisp, and interdental tongue position for swallowing	Group A: traditional articulation treatment. Group B: tongue thrust treatment incorporating neuromuscular facilitation techniques for first 6wks followed by alternating sessions of tongue thrust and articulation treatment for the next 8wks.	Participants received a total of 22 individual 0.5-h treatment sessions Therapy was provided once a week for 6wks followed by twice a week for 8wks.	Three-point scale for determining tongue thrust severity in swallowing of solids, liquids, and saliva. Group B significant gains compared with group A	$p<0.05$	NR	5/8
Ganz[28]	1	8:0	1 female	CP and profound mental* retardation with protraction and extension of tongue during feeding and tonic bite reflex.	A-B-A design with oral-motor intervention program of manual vibration, stretch pressure, gum massage, and pressure application to teeth and tongue.	18-wk study: phase 1: 2wks baseline; phase 2: 14wks of intervention. Treatment provided five times per week during mealtime. Phase 3: 2wks return to baseline.	Decreased frequency of: tongue thrusting with solid foods, phase 1–2	$p<0.001$	NR	3/7
							Increased frequency of tongue thrusting with solid foods, phase 2–3	$p<0.04$		
							Decreased tongue thrusting with semi-solid foods, phase 1–2	$p<0.001$		
							Frequency of tongue thrusting with semi-solid foods, phase 2–3	NS		
							Frequency of tonic bite, phase 1–2	NS		
Gisel et al.[29]	1	12:0	1 female	CP with moderate spastic quadriparesis and psychomotor retardation	ISMAR- an intra-oral stimulating plate.	After a 6-mo no treatment baseline, the appliance was worn for 12mo. Frequency and intensity were not reported.	FFA	NR	NR	2/7

Table II: Continued

Reference	*n*	Age in y:mo (range and/or mean)	Sex	Medical and/or speech-language pathology diagnosis	Intervention	Treatment schedule and amount	Outcome measure(s) and findings		Effect size (95% confidence interval)	Quality marker score
							Amount of time to take five bites of three standard textures	NR	NR	
Gisel et al.[31]	17	6:7–15:5 (10:2)	7 males, 10 females	Spastic CP with moderate dysphagia	Group A: ISMAR an intra-oral stimulating plate. Group B: standard rehabilitation at school and delayed treatment with intra-oral stimulating plate.	Daily wear for 12mo	FFA: chewing domain from month 12 to 24			3/8
							Intervention group	NS	−0.09 (−0.99, 0.81)	
							Comparison group	NS	0.14 (−0.88, 1.15)	
							FFA: swallowing domain from month 12 to 24			
							Intervention group	NS	−0.07 (−0.97, 0.83)	
							Comparison group	NS	0.48 (−0.57, 1.48)	
							FFA: clearing domain from month 12 24			
							Intervention group	NS	0.17 (−0.74, 1.07)	
							Comparison group	NS	0.57 (−0.49, 1.57)	
Haberfellner et al.[32]	20	4–13 (8:4)	9 males, 11 females	Spastic CP	Group A: Innsbruck sensorimotor activator and regulator, an intra-oral stimulating plate. Group B: Standard rehabilitation at school and delayed treatment with intra-oral stimulating plate.	Group A: no-treatment period 6mo. Phase 1 treatment period to stabilize mandible: 6mo. Phase 2 treatment period to facilitate ingestive skills: 6mo. Group B: NR.	FFA: chewing domain			4/8
							Group A	$p<0.001$	1.99 (0.85, 2.96)	
							Group B	NS	0.27 (−1.14, 0.62)	
							FFA: swallowing domain			
							Group A	NS	0.66 (−0.26, 1.53)	
							Group B	NS	0.19 (−0.7, 1.06)	
							FFA: clearing domain			
							Group A	NS	0.20 (−0.69, 1.07)	
							Group B	NS	0.15 (−0.73, 1.02)	
Harden & Rydell[33]	80: intervention group=50; comparison group=30	13–32 (16:5)	Intervention group 27 males, 23 females Comparison group 17 males, 13 females	Tongue thrust during swallowing	Intervention group: tongue thrust treatment. Comparison group: no treatment.	Evaluated children for outcomes of treatment that had been completed at least 5y before. No information on original treatment schedule reported.	Three-point tongue thrust severity rating scale	$p<0.001$	NR	3/8

inued

n	Age in y:mo (range and/or mean)	Sex	Medical and/or speech-language pathology diagnosis	Intervention	Treatment schedule and amount	Outcome measure(s) and findings		Effect size (95% confidence interval)	Quality marker score
45: intervention group=26; comparison group=19	3:11–16:11 (8:4)	32 males, 13 females	Multiple untreated orofacial dysfunctions	Intervention group: face former therapy which consisted of a series of lip and tongue exercises with a flexible silicone training device. Comparison group: conventional myofunctional therapy	Both groups were followed for 6mo. Intervention group performed 20 repetitions of the exercises three times per day. After 3wks the training device was worn overnight. Comparison group: not reported.	Clinical examination by palatography	χ^2 test at 6mo: p=0.028	NR	4/8
4	3–12 (6:6)	1 male, 3 females	Severe multiple disabilities	A-B-A-B reversal experimental design. Oral and perioral stimulation including stroking lips and cheeks with soft cloth, rubbing hard palate and gums, applying ice to lips and cheeks, stretch-pressure to lips and cheeks, and perioral and facial vibration and brushing.	Facilitation procedures provided for 10–15min before lunch at school.	Clinical observation of percentage of bites with lip closure, percentage of chews with rotary pattern, number of spills per bite, and number of spills per drink.	NR	NR	2/7

ning disability. FFA, Functional Feeding Assessment; NR, not reported or calculable; NS, not significant; CP, cerebral palsy; ISMAR, Innsbruck Sensorimotor Activator and Regulator.

Three studies [29,31,32] by the same group investigated the impact of the Innsbruck Sensorimotor Activator and Regulator (ISMAR), an intra-oral stimulating appliance, on swallowing physiology outcomes measured by the Functional Feeding Assessment (FFA)[40] in children with CP. Haberfellner et al.[32] compared the use of OME (i.e. ISMAR) to standard rehabilitation received at school. Gisel et al.[31] evaluated children who continued to wear the ISMAR for a second year compared with those who did not. The other study by Gisel et al.29 used a single-subject design to compare ISMAR usage with a 6-month no-treatment baseline phase.

Haberfellner et al.[32] reported that the ISMAR had a large positive effect (d=1.99) on the chewing domain of the FFA. Smaller effects were noted on the swallowing domain (d=0.66) and the clearing domain (d=0.2) but these were not statistically significant. The group receiving standard rehabilitation at school demonstrated no significant changes on the chewing domain (d=0.27), swallowing domain (d=0.19), or the clearing domain (d=0.15) of the FFA. Gisel et al.[31] continued to track these two groups for 12 months and re-assessed their performance on the FFA. At the 1-year follow-up, the group that continued to wear the ISMAR device exhibited no change on the chewing domain (d=−0.09), swallowing domain (d=−0.07), or clearing domain (d=0.17) of the FFA. The group who discontinued use of the ISMAR also showed no change on the chewing domain (d=0.14) of the FFA. However, some changes were noted on the swallowing domain (d=0.48) and the clearing domain (d=0.57), but these were not statistically significant. These authors concluded that during the 1-year follow-up period, previous treatment gains were maintained and that maturation alone was equally as effective as ISMAR treatment. The third study29 also investigated the use of the ISMAR but did not provide sufficient information to analyse the findings.

Three studies [26,33,36] examined the impact of tongue thrust treatment or lip and tongue exercises on the swallowing physiology skills in children. None of the studies provided sufficient data to calculate effect sizes. Christensen and Hanson26 reported that children receiving OME plus articulation treatment made greater gains ($p<0.05$) than those receiving articulation treatment alone on a three-point scale of tongue thrust severity. Harden and Rydell[33] found that the group receiving tongue thrust treatment performed significantly better ($p<0.001$) than a no-treatment comparison group on a three-point tongue-thrust severity rating scale 5 years after treatment. Korbmacher et al.[36] compared two different forms of OME. They noted that children who participated in Face Former treatment (a series of lip and tongue exercises with a flexible silicone training device that is inserted behind the lips but in front of the teeth) exhibited significantly greater improvement in swallowing patterns (p=0.028) than those who received conventional myofunctional treatment.

Two studies [28,39] used a single-subject design to examine the effects of oral, perioral, and facial stimulation on the swallowing physiology skills of children with multiple disabilities. However, only Ganz28 reported adequate data to analyse the findings statistically. Significant differences were noted between the baseline and treatment

phases for decreased tongue thrusting with solid foods ($p<0.001$) and semi-solid foods ($p<0.001$). As a follow-up to determine if treatment effects were maintained once treatment was discontinued, the frequency of tongue thrust was measured during a 2-week return to baseline phase. During this phase, a significant increase in tongue thrusting was observed with solid foods ($p<0.04$) but not with semi-solid foods. No significant difference was observed in the frequency of tonic bites during oral feeding with treatment.

Table SII (published online only) reports the methodological quality ratings for the studies examining swallowing physiology outcomes. Quality marker scores for the five controlled trials[26,31–33,36] ranged from three to five out of a possible score of eight. The scores for the three single-subject design studies[28,29,39] ranged from two to three out of a possible seven. Most studies (6/8) provided information about the statistical significance of the findings.[26,28,31–33,36] However, several methodological weaknesses were noted. Half of the studies (4/8) reported assessor blinding,[26,29,32,36] used validated outcome measures,[26,28,33,39] or provided an adequate description of the participants or group comparability.[28,29,36,39] Only two of the eight studies provided sufficient data to calculate effect sizes and confidence intervals,[31,32] and only one of the controlled trials used an intention-to-treat standard in data analysis.[26] None of the studies reported random allocation of participants or provided an adequate description of randomization procedures.

Clinical question 2: what is the effect of OME on pulmonary health in children?

No study was found to address this clinical question.

Clinical question 3: what is the effect of OME on functional swallowing outcomes in children?

Table III provides a detailed description of the intervention and participants reported in the six studies that addressed the effectiveness of OME on functional swallowing outcomes in children. Four studies examined the effects of an intra-oral stimulating appliance and two studies evaluated the use of oral stimulation and sensorimotor facilitation procedures.

Table III: Functional swallowing outcomes (question 3) summary table

Reference	*n*	Age in y:mo (range and/or mean)	Sex	Medical and/or Speech Language Pathologist diagnosis	Intervention	Treatment schedule and amount	Outcome measure(s) and findings		Effect size (95% confidence interval)	Quality marker score
Gisel et al.[29]	1	12	1 female	CP with moderate spastic quadraparesis and psychomotor retardation	ISMAR, an intra-oral stimulating plate.	The plate was worn for 12mo. Frequency and intensity were not reported.	Weight gain	NR	NR	3/7
Gisel et al.[30]	20	4–13 (8:4)	11 males, 9 females	Spastic CP	Group A: ISMAR, an intra-oral stimulating plate. Group B: standard rehabilitation at school and delayed treatment with intra-oral stimulating plate.	Participants were instructed to wear the plate at night during the two treatment phases. Each treatment phase lasted 6mo.	Changes in diet level/food textures	NR	NR	1/8
Gisel et al.[31]	17	6:7–15:5 (10:2)	7 males, 10 females	Spastic CP with moderate dysphagia	Intervention group: Innsbruck sensorimotor activator and regulator, an intra-oral stimulating plate. Comparison group: no treatment.	NR	Change in weight (kg) Change in height (cm)	NS NS	0.22 (−0.76, 1.18) 1.05 (−0.03, 2.01)	4/8
Haberfellner, et al.[32]	20	4–13 (8:4)	11 males, 9 females	Spastic CP	Group A: ISMAR an intra-oral stimulating plate. Group B: standard rehabilitation at school and delayed treatment with intra-oral stimulating plate.	Participants were instructed to wear the plate at night during the two treatment phases. Each treatment phase lasted 6mo.	Weight gain	NS	NR	4/8
Ottenbacher, et al.[14]	20	5–21 (11:6)	12 males, 8 females	All participants had severe to profound mental retardation* and some degree of neuromotor disorders. Eighteen of the participants had an additional diagnosis of CP	Intervention group: sensorimotor facilitation procedures to normalize tone, reduce abnormal reflexes, and desensitize oral region. Comparison group: no treatment.	Treatment was administered 30–40min a day, 5d a week, for 9wks.	Weight	NS	0.58	5/8
Ottenbacher, et al.[37]	3	8–12 (10:0)	NR	Severe to profound mental retardation and some degree of neuromotor disorder	Sensorimotor facilitation procedures to normalize tone and reduce abnormal reflexes.	Treatment was scheduled for 30min a day, 5d/wk over 12wks.	Participant 1: weight loss Participant 2: weight gain Participant 4: weight change	p=0.001 p=0.004 NS	NR	3/7

*UK usage: learning disability. NR, not reported or calculable; NS, not significant; CP, cerebral palsy; ISMAR, Innsbruck Sensorimotor Activator and Regulator.

Four studies investigated the use of the ISMAR on functional swallowing outcomes.[29–32] A total of 58 participants (age range 4y 5mo–15y 5mo) with a diagnosis of spastic CP were included in the data analyses.

In three studies, participants used the ISMAR for a 12-month treatment period.[29,30,32] Treatment consisted of two 6-month phases. The first phase targeted jaw stabilization, the second phase targeted oral structure mobilization. In the single-subject design study, the jaw stabilization phase served as the control for the one child.[29] These studies reported the impact of the intra-oral stimulating plate on functional swallowing outcomes, including weight gain [29,31] and change in diet level.30 However, only one study reported adequate data to analyse the findings statistically.[32] No between group difference was found in weight gain. A final study investigated the long-term effects of OME on weight gain and growth.31 As a follow-up to Haberfellner et al.,32 this study tracked participants who continued to wear the ISMAR for an additional year compared with those who did not and found that OMEs had no significant effect on weight gain (d=0.22) or growth (d=1.05) after 1 year.

Two studies examined the effects of oral stimulation and neuromuscular facilitation procedures on weight gain in young people with severe to profound mental retardation† and neuromotor disorders.[14,37] One reported that OME had an effect on weight gain (d=0.58) in this population. However, the difference between the intervention and comparison groups was not statistically significant.[14] The other reported significant weight gain for one participant, significant weight loss for another, and no change in weight for a third participant.[37]

Table SIII (published online only) reports the methodological quality ratings for studies addressing this question. Quality markers ranged from one to five out of eight possible markers for the four controlled trials.[14,30–32] The two single-subject design studies both received a total of three out of seven possible markers.[29,37] Most of these studies provided valid and reliable outcome measures,[14,29,31,32,37] blinded the assessors to the treatment condition,[14,29,32,37] and reported measures of statistical significance.[14,31,32,37] Only one study provided effect size and confidence interval data.[31] Methodological weaknesses for the included studies were lack of randomization, group/participant comparability, and intention-to-treat analysis when appropriate.

Clinical question 4: what is the effect of OME on drooling management in children?

The five studies that provided data to address the effects of OME in children with drooling issues are listed in Table IV.[25,27,34,35,38] Three studies [27,35,38] examined the use of oral stimulation and facilitation in children and young adults with CP and one study [34] investigated the use of chin cup intervention and OME classes in children and adolescents with CP. The fifth study evaluated the use of oral stimulating plates in children with Down syndrome.[25]

Table IV: Question 4: Drooling management summary

Reference	*n*	Age in y:mo (range and/or mean)	Sex	Medical and/or Speech Language Pathologist diagnosis	Intervention	Treatment schedule and amount	Outcome measure(s) and findings		Effect size (95% confidence interval)	Quality marker score
Calrlstedt et al.[25]	20	(5:7)	12 males, 8 females	Down syndrome	Intervention group- oral stimulating plate therapy plus physiotherapy program designed by SLP. Comparison group: physiotherapy program designed by SLP.	Plates worn for 1h two or three times per day for a minimum of 4y (range, 49–58mo). Control group: not reported.	Parent assessment of drooling using visual analog scale (0–100points) Drooling during day Drooling at night	 NS NS	 0.19 (–1.07, 0.7) 0.45 (–0.46, 1.32)	3/8
Domaracki & Sisson[27]	2	(10:0)	1 male, 1 female	CP and severe/profound mental retardation	Oral stimulation of hard palate, gums, tongue, and cheeks. Use of rounded toothbrush with soft rubber bristles and stimulation with adaptability two-speed vibrator.	Hourly treatment of oral stimulation to hard palate, gums, tongue and cheeks (five to ten reps each). 10-s stimulation to chin and throat with vibrator.	Percentage of drooling intervals	NR	NR	1/7
Harris & Dignam[34]	20	6-15 (9:8)	9 males, 11 females	CP. Moderate to profuse drooling.	Group 1: chin cup intervention plus oral motor exercise class. Group 2: chin cup intervention plus oral motor exercise class. Group 3: oral motor exercise class only. Group 4: No treatment.	Chin cup 6h five times per week. OME class 30-min sessions three to five times per week. Chin cup duration: 3–6mo. OME class: 9–14mo.	Percentage of drooling by bib weight	NR	NR	1/8
Iammatteo et al.[35]	2	S1=2:7 S2=2:11	2 males	CP	Oral facilitation (firm pressure to lips and mouth before feeding, and jaw and lip control during feeding).	30-min sessions/12d	Pre/post-saliva bib weight Participant 1 Participant 2	 NS Stated statistically significant	 NR NR	3/7
Samelstad[38]	2	S1=14:0 S2=21:0	2 females	Mental retardation* and CP	Facilitation of lip closure and swallowing through stroking and stretching, and pressure around lips and larynx.	37–38 sessions Once per day, 5d/wk	Pre/post-bib saliva weight	NR	NR	1/7

*UK usage: learning disability. NR, not reported or calculable; NS, not significant.

Cohen's d values were calculable for one study.[25] OME plus oral stimulating plates had a negligible effect (d=0.19) compared with OME alone on parental perception of daytime drooling and a small effect (d=0.45) on night time drooling. Iammatteo et al.[35] provided additional data for this clinical question but no effect sizes were calculable in that report. OME had a significant effect on pre-/post-saliva bib weight for one participant but not for the other. The remaining studies did not provide sufficient data to analyse the findings.

Table SIV (published online only) shows the methodological quality ratings for each study. Two of the five studies were controlled trials so all eight quality markers applied.[25,34] The remaining three were considered single-subject designs so the eighth marker (intent-to-treat analysis) was not relevant.[27,35,38] Two of the single-subject design studies provided an adequate description of included participants [35,38] and two used validated outcome measures.[27,35] Only two studies provided information about the statistical significance of the findings.[25,35] Methodological weaknesses were also apparent. None of the studies reported blinded assessment. Neither random allocation of participants nor adequate description of randomization procedures was reported in any of these studies. Moreover, none of the controlled trials reported using an intention-to-treat standard in data analysis.

Effect of study quality on results

The results of the included studies were examined to ascertain if differences in methodological quality were associated with differences in effect sizes. For each clinical question, the magnitude of the effect sizes was investigated under different methodological conditions to determine the impact of an individual quality marker on overall study results. However, because so few effect sizes were reported or calculable, and there were only minimal discrepancies among the included studies in quality markers, no conclusions were possible.

DISCUSSION

The aim of this EBSR was to determine the effects of OME on physiological and functional swallowing outcomes, pulmonary health, and drooling management in children. Overall, the findings showed limited support, at best, for the narrow application of some specific OME treatments and no support for others. A systematic search of the scientific literature yielded 16 studies, with considerable methodological limitations, that addressed three of the four clinical questions. No study was found to address effect of OME on pulmonary health in children. Thirty-six findings were reported across the 16 studies. Of these, 28 could be analysed statistically either through the reporting or calculation of effect sizes and/or statistical significance. Mixed results were noted across these 28 outcome measures, with OME resulting in positive changes on swallowing skills or drooling on 12 of the outcome measures, no change (or negative change, noted in one study[37]) on 11

measures, maintenance of treatment effects on four measures, and no maintenance of previous treatment gains on one measure. Within the 12 positive findings, four outcomes had effect sizes ranging from 0.45 to 1.05 but the differences were not statistically significant. The lack of statistical significance may be a result of the small sample sizes in these studies or to the large variance of the results, although it is not possible to provide one specific reason or cause. The largest and most notable positive treatment effect (d=1.99) was reported for the initial 12-month use of the ISMAR oral stimulating plate in children with CP on the chewing domain of the FFA.[32] However, the importance of this finding is questionable because at a 1-year follow-up, the group that continued to wear the ISMAR for a second year showed comparable results to those who discontinued ISMAR use. This indicates that maturation alone was equally as effective as the ISMAR during that timeframe. The results of this EBSR highlight that OMEs are not a unitary phenomenon. There are many variations in applications of OME across multiple populations of children with different etiologies, ages, degrees of deficit, and risk factors for pharyngeal swallow problems that may include aspiration with oral feeding and on saliva/secretions. Hence, it is not surprising that mixed results were found.

Most studies (10/16) in this EBSR investigated the use of OME in children with CP, who make up a very diverse population.[41] Participants in the included studies probably reflected wide heterogeneity that could account for some of the inconsistency of the findings. Moreover, only half of the studies examining this population provided an adequate description of the participants or reported group comparability at baseline. Given the importance of understanding how individual variables may influence treatment outcomes, provision of detailed and thorough descriptions of study participants is a requisite component to interpreting the results of individual studies as well as systematic reviews.

Another factor contributing to these mixed results may be the wide variety of OME incorporated into interventions used across studies that included children with CP. Because OME were defined broadly in this EBSR, many different and isparate types of intervention were examined, including oral stimulating plates, sensorimotor facilitation, oral-motor exercise classes, and sensory input involving massage, stretching, vibration, stroking, or pressure application to the oral or perioral regions. Often these interventions were not used in isolation, but instead in combination with other OME (e.g. massage used with vibration and stroking). In clinical contexts it is reasonable to consider multiple or combinations of interventions with children who have such complex interrelated deficits. However, it is difficult to impossible to investigate the impact of this approach or to identify the active ingredient(s) that may have contributed to or inhibited the desired outcome when these interventions are combined.

More consistent findings were noted for the use of OME in children who exhibited a tongue thrusting pattern during swallowing but who did not have a concomitant defined neurological diagnosis. Although none of the studies that focused on tongue thrust treatment provided sufficient data to calculate effect sizes, all three reported

that OME had a statistically significant effect on swallowing physiology compared with articulation treatment,[26] no treatment,[33] or another form of OME, specifically conventional myofunctional treatment.[36] Although each study had some methodological weaknesses, these limited results are promising for providing a framework for future research. Similarly, the definitions of quality indicators used as a basis for this systematic review (Table I) should be of assistance to investigators in the future to improve the quality of their study designs and methodologies. It is hoped that future studies will have increased impact and credibility compared with the current state of the evidence with use of OME for these areas of intervention.

Implications for clinical practice

Because the results from the studies were mixed, the study participants were heterogeneous and not consistently described, and the included studies incorporated a variety of interventions, this EBSR may pose more questions than it answers. In addition, the equivocal results highlight the clinical uncertainty underlying the use of these interventions and do little to settle the debates and controversies surrounding OME.

When evidence-based data are not available, or when outcomes are contradictory, clinicians must rely on their knowledge of anatomy, physiology, and neurodevelopment to address oral sensorimotor deficits and swallowing problems (dysphagia). Clinicians need to understand typical neurodevelopment to understand differences in neurodevelopment, particularly as they consider intervention strategies in children with neurological damage, as is the case in all children with CP, Down syndrome, and several genetic syndromes and craniofacial anomalies. Principles of experience-dependent neural plasticity provide a basis for learning as a primary means for remodeling the damaged brain,[42] regardless of when the brain damage occurs, for example in utero, during the neonatal period, or at any time in life. Although not specific to children with dysphagia, these principles of motor learning can provide guidance to clinicians in selecting appropriate therapeutic strategies when only limited or equivocal research is available. For example, as discussed previously, the principle of specificity suggests that a treatment exercise should closely parallel the desired task. Two other principles, 'age matters' and 'time matters', have implications for the timing of intervention. Given that neural plasticity decreases as aging occurs, younger children are likely to be more responsive to training-induced changes in neural function. Furthermore, the principle of 'time matters' suggests that treatment initiated earlier in the injury-recovery process may increase neural sensitivity to the effects of behavioral experience. Although clinical evidence is not yet available for these principles, they do provide a reasonable rationale for clinicians. However, given the lack of treatment evidence, clinicians should take steps to evaluate carefully the effects of these exploratory treatments within a controlled treatment design.

Limitations of the current review

Several limitations should be considered when interpreting the results of this EBSR. First, only articles published in English were considered for inclusion. Therefore, it is possible that some relevant studies in other languages were not identified. Second, only studies published in the peer-reviewed literature were included. This exclusion could have introduced publication bias or the over-representation of positive treatment effects into the results of this EBSR. Additionally, OME was defined broadly in this review, thereby including a wide range of interventions. Although the breadth of this definition was deliberate to capture the broad scope of interventions clinicians consider to be within the realm of OME and use in clinical practice, it also introduced variability that restricted our ability to combine or compare results across studies. Another limitation that precluded us from comparing results across studies was that few of the included studies provided adequate or detailed descriptions of the interventions to allow for these comparisons or analyses. Finally, each of the included studies failed to meet at least three of the quality indicators (out of a possible seven or eight) that were assessed as part of the quality appraisal process. Many of the common methodological shortcomings identified (e.g. lack of assessor blinding, data not analysed by an intention-to-treat protocol, inadequate allocation) have been associated with inflating treatment effects43 and therefore limiting the confidence clinicians can have in the findings. This systematic review should be considered current as of August 2007. Any relevant studies published after this date were not included. Because new studies continue to emerge about the effects of OME on swallowing and drooling in children, clinicians are encouraged to re-examine the available evidence on this topic regularly.

The results of this EBSR elucidated several key areas for future research. Because no study was found that examined the effects of OME on pulmonary health in children, well-designed and highly controlled investigations are still needed for this important clinical outcome. Few studies provided sufficient data on the effects of OME on drooling. Given that drooling may be stressful or socially isolating for children, additional studies are needed to determine effective treatment options. Furthermore, to determine which individual child characteristics may influence treatment outcomes, future research should include detailed descriptions of participants and treatment protocols. Maintenance of treatment effects was explored in only two studies.[28,31] However, given the mixed findings and limited types of OME examined, additional investigations are necessary. Research is needed in all aspects of intervention programs for children with oral sensorimotor deficits. It is important to define the population(s), determine measurable goals, take into account expected changes by maturation, reduce variables, and overcome the methodological limitations noted in the studies that met criteria for inclusion in this review of OME in children. This review of the literature was conducted to determine whether support for OME could be found in reports of studied treatment strategies. It was hypothesized that findings would aid clinicians in their daily practice and could form a basis for research into unstudied strategies. However, these equivocal

findings provide no definitive evidence for the use of OME in daily practice. Decision-making strategies for clinical practice typically involve knowledge of evidence-based research reports that describe the benefits of a particular treatment when those reports are available. When evidence is not available, clinicians use their knowledge on the basis of theoretical soundness of an approach. This approach requires extensive knowledge of how common neuromuscular dysfunctions affect movement or how motor-based treatments act to influence underlying impairments.[12] Selection of techniques will be difficult for clinicians who do not have that extensive knowledge of the neurophysiological bases of neuromuscular dysfunctions. It is critical that clinicians can evaluate information in this area to provide interventions that will result in positive functional outcomes for their patients. It is hoped that all clinicians involved in the care of these children examine the evidence on which interventions are based as well as develop appropriate research studies to contribute to the literature.

What this paper adds

- The first evidence-based systematic review examining the effects of oral motor exercise on swallowing physiology, pulmonary health, functional swallowing, and drooling management in children.

- Comprehensive quality appraisal and summary of 16 included studies.
- No definitive findings to support treatment of swallowing problems with oral motor exercise.

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