Oral Sensory-Motor Intervention for Children and Adolescents (3-18 Years) With Dysphagia or Impaired Saliva Control Secondary to Congenital or Early-Acquired Disabilities: A Review of the Literature, 2000 to 2016

Lotta Sjögreen, PhD1, Margareta Gonzalez Lindh2, Madeleine Brodén, MS3, Corinna Krüssenberg, MS4, Irvina Ristic, MS5, Agneta Rubensson, MS6, and Anita McAllister, PhD7

Abstract
Objectives: Dysphagia and impaired saliva control are common in children and adolescents with congenital and developmental disabilities. The aim of the present review was to investigate the evidence base for oral sensory-motor interventions in children and adolescents with dysphagia or impaired saliva control secondary to congenital or early-acquired disabilities and to make recommendations regarding methods for intervention.

Methods: A review of the literature from 2000 to 2016, including oral sensory-motor intervention studies for children and adolescents (3-18 years of age) with dysphagia or impaired saliva control secondary to congenital or early-acquired disabilities, was performed. The literature search included the PubMed, CINAHL, Medline, SpeechBITE, OVID, ERIC, Cochrane, and Google Scholar databases. Primary studies were evaluated on a 4-grade scale using the Grading of Recommendations Assessment, Development and Evaluation.

Results: Twenty primary studies of oral sensory-motor interventions for dysphagia and 6 studies for the treatment of impaired saliva control fulfilled the inclusion criteria. Of these, 3 were randomized, controlled trials. Five systematic reviews and 16 narrative reviews were also included. Limited and moderately strong recommendations were made on the basis of the grading results from the primary studies. The studies reported good results, but study design was often insufficient, and the study groups were small. The systematic reviews confirmed the lack of high scientific support for oral sensory-motor interventions in children and adolescents with congenital and developmental disabilities.

Conclusions: There is an urgent need for high-quality studies that could serve as the basis for strong recommendations relating to oral sensory-motor interventions for children with dysphagia and impaired saliva control.

Keywords: deglutition, deglutition disorders, children, intervention, drooling, dysphagia

Introduction
Oral sensory-motor dysfunction in children and adolescents with congenital and developmental disabilities often affects swallowing and causes dysphagia and impaired saliva control (sialorrhea). Feeding difficulties in infants can be caused by abnormality of the organs or failure of the appropriate maturation of functions and reflexes. Furthermore, the cause of swallowing problems can be either organic or behavioral or both, but, regardless of the cause, feeding development will probably be influenced.1,2

Dysphagia can occur with solids and/or liquids and is categorized as oropharyngeal or esophageal. In young or developmentally disabled children, the symptoms can be subtle, manifesting only as slow eating, multiple swallowing attempts, or aspiration pneumonia, or obvious.
manifesting as coughing, choking, or vomiting. In cases of severe dysphagia, there is an increased risk for life-threatening events such as choking, aspiration, and malnutrition.3

Impaired saliva control is often a consequence of either poor oral muscle tone or dysphagia. Some studies suggest a correlation between decreased oral sensation and saliva leakage.4 Saliva leakage, chewing with an open mouth, and coughing during meals are often stigmatizing and can exclude individuals from participating in meals and social interaction.

Oral sensory-motor therapy uses a variety of exercises to develop awareness, strength, coordination and the movement and endurance of the lips, cheeks, tongue, and jaw. They include active muscle exercise, muscle stretching, passive exercise, and sensory stimulation and aim to influence the physiology of the oropharyngeal mechanism.5,6

The ways parents of children with disabilities experience their children’s oral sensory-motor dysfunction have been identified through interviews and questionnaires, and guardians often say that these dysfunctions have a considerable influence on quality of life.7,9 In a study (the Feeding/Swallowing Impact Survey), Lefton-Greif et al10 identified 3 main problem areas reported by the parents of children with dysphagia: daily activities, anxiety, and eating difficulties. On the basis of the health-related and social effects of dysphagia and impaired saliva control, it is important to make evidence-based interventions available to this patient group.

Before any intervention can be recommended, many questions need to be considered and discussed with the patient and caregiver: Is it likely that the recommended intervention will lead to the intended result? Is the recommended intervention better and more effective than other methods? Does the patient have the necessary resources (including support) and enough motivation and strength to carry out the intervention? To be able to answer these and similar questions, the clinician needs knowledge of the evidence base for different and alternative interventions for a specific dysfunction in a certain patient group. The final decision on the method that is best suited to the individual patient is made by the patient or a caregiver after having been informed of the possible alternatives.

The implementation of new research often takes time because of a variety of personal and administrative obstacles, such as a lack of researchers in an organization, a lack of resources, or an inborn resistance to reorganization.11 Introducing new methods for intervention is expensive and takes time and resources from ordinary clinical work. It is therefore of great importance that the chosen intervention be the most effective for the specific patient group.

The aim of the present review was to investigate the evidence base for oral sensory-motor interventions in children and adolescents aged 3 to 18 years with dysphagia or impaired saliva control secondary to congenital or early-acquired disabilities. On the basis of the literature review, another aim was to make recommendations regarding different therapy methods for improving or preserving oral sensory-motor functions in children and adolescents with dysphagia or impaired saliva control secondary to congenital or early-acquired disabilities.

Methods

The inclusion criteria were as follows: (1) articles published in scientific peer-reviewed journals between January 1, 2000, and February 29, 2016; (2) children from the age of 3 years to adulthood; (3) a diagnosis of congenital or developmental disability; and (4) reported speech and oral sensory-motor dysfunction, including swallowing dysfunction, drooling, and oral habits. The present review was part of a larger project that also included an evaluation of oral sensory-motor interventions for speech.12

A literature search was performed using the PubMed, CINAHL, Medline, SpeechBITE, OVID, ERIC, Cochrane, and Google Scholar databases. The search was limited to intervention studies published in English, Swedish, or German between January 1, 2000, and February 29, 2016. Articles that fulfilled the inclusion criteria and were identified during the reading process were added to the project.

The Grading of Recommendations Assessment, Development and Evaluation (GRADE) was used to make a qualitative evaluation of the primary studies that were reviewed.13 The grading system uses a 4-grade scale for an intervention method: 4 = high, 3 = moderate, 2 = limited, and 1 = insufficient quality. The study design was crucial to the grading. Highly ranked study designs were randomized controlled trials, meta-analyses, and systematic reviews (GRADE 4). Controlled trials were evaluated as being of moderate quality (GRADE 3). Observational studies without controls and studies with multiple single-subject designs were considered to be of limited quality (GRADE 2), and case reports were judged to be of insufficient quality (GRADE 1). In addition, a study could be upgraded or downgraded depending on how it was performed and reported and the representativeness of the study population.14

The final recommendations for an intervention method for a specific patient population were based on the results of the GRADE evaluation. The following recommendations were used:

**High scientific support**: The method was used in several studies with high or medium to high quality and no or few diminishing factors.

**Medium to high scientific support**: Methods with high or medium to high quality with diminishing factors in the overall assessment of the assessed studies.
Limited scientific support: Methods with high or medium to high quality and substantial diminishing factors in the overall assessment of the assessed studies.

Insufficient scientific support: Methods for which the number of studies was few or of low quality or the available studies reported conflicting results.

Results

Initially, 3,555 articles were found, 1,131 about dysphagia and 2,424 about saliva leakage (Figure 1). After the deletion of all duplicates and obviously irrelevant articles, abstracts from the remaining articles were read and categorized according to participants, diagnosis groups, and types of interventions. Only studies that related to oral sensory-motor interventions and included participants with congenital and early-acquired disabilities were finally selected for the review. Five systematic reviews, 16 narrative reviews, and 26 primary studies, of which 3 were randomized controlled trials, were finally included in the analysis. A variety of methods for the treatment of dysphagia (Table 1) and saliva leakage (Table 2) were found.

In the original project (including also interventions for speech), a total of 28 articles (28.5%) were randomly selected for interrater reliability testing. The selected studies were independently graded by 2 investigators. The interrater reliability proved to be very good (Cronbach α = .986).

Interventions for Children and Adolescents With Dysphagia Secondary to Congenital or Early-Acquired Disabilities

Primary Studies. Twenty primary studies of oral sensory-motor interventions for children and adolescents with dysphagia secondary to congenital or early-acquired disabilities fulfilled the inclusion criteria (Table 1). In all, 556 children and adolescents aged 0.4 to 17 years participated in these studies. Twenty children with Down syndrome were involved in 2 studies. The included diagnoses were cerebral palsy (n = 86),17-20 Down syndrome (n = 50),15,16,21-23 myotonic dystrophy type 1 (n = 8),24 Goldenhar syndrome (n = 3),25 food refusal (n = 45),26 deviant swallowing pattern (n = 157),27-29 oromotor dysfunction (n = 45),30 and dysphagia of unspecified etiology (n = 157).31-34 Oral sensory-motor stimulation was often part of a more holistic treatment program also including feeding and swallowing therapy, mealtime adaptations, and behavior management.

Twenty children with cerebral palsy and moderate dysphagia were treated with the Innsbruck Sensorimotor Activator and Regulator (ISMAR) for 1 year. Ten of the children were randomly chosen to act as control subjects and entered the therapy program at a later stage. The abilities to spoon-feed, bite, chew, and drink from a cup improved during the intervention period. The improvements were significant compared with what could be expected from spontaneous development only. In a
follow-up study, no difference was found between the group of children who were treated for 2 years and the control group, which finished the treatment after 1 year.¹⁸

Five intervention studies for children with food refusal problems comprised a total of 119 children, aged 4 months to 15 years. The therapy focused on oral tactile stimulation, but it was often combined with other interventions, such as swallowing therapy and behavior modification.²¹,²⁵,²⁶,³²,³³ In 1 of the studies, 45 children with severe dysphagia were treated with intraoral tactile stimulation.²⁶ Before therapy, they were all dependent on artificial nutrition. At follow-up, 1 to 2 years later, nearly all of them (93%) were able to eat complete meals orally. In all studies in which the intervention of oral tactile stimulation was included, the children made significant improvements. None of the studies included controls, but the concurrent results point in the

<table>
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<tr>
<th>Table 1. Intervention Studies Including Individuals With Dysphagia Caused by Congenital or Early-Acquired Disabilities, Type of Intervention, Number of Participants, and Grading.</th>
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<tbody>
<tr>
<td><strong>Participants</strong></td>
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<tr>
<td>Reference</td>
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<tr>
<td>Haberfellner et al (2001)¹⁷</td>
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<td>Carlstedt et al (2003)¹⁶</td>
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<td>Carlstedt et al (2001)¹⁵</td>
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<td>Schwarz et al (2001)²⁴</td>
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<td>Lamm et al (2005)²⁶</td>
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<td>Korbmacher et al (2004)³⁰</td>
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<td>Smithpeter and Covell (2010)²¹</td>
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<td>Eckman et al (2008)²³</td>
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<td>Clawson et al (2007)²⁰</td>
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<td>Bailey and Angell (2005)³²</td>
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<td>Tamura et al (2011)²⁵</td>
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<td>Clawson et al (2006)²⁵</td>
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<td>Saccomanno et al (2012)²⁷</td>
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<td>Green (2013)³⁸</td>
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<td>Giuca et al (2008)²⁹</td>
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Abbreviations: GRADE = Grading of Recommendations Assessment, Development and Evaluation; ISMAR = Innsbruck Sensorimotor Activator and Regulator; OMT = oral myofunctional therapy.

*Part of function as outcome.*

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<tr>
<th>Table 2. Intervention Studies Using Intra-Oral Appliances Including Children and Adolescents With Impaired Saliva Control (Drooling) Caused by Congenital or Early-Acquired Disabilities, Number of Included Participants, Diagnoses, and Grading.</th>
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<tbody>
<tr>
<td><strong>Participants</strong></td>
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<tr>
<td>Reference</td>
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<td>Alacam and Kolcuoğlu (2007)¹⁹</td>
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<td>Gerek &amp; Çiyiltepe (2005)⁵¹</td>
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<tr>
<td>Koskimies et al (2011)⁹⁹</td>
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<td>Pani and Hegde (2007)⁵³</td>
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<td>Johnson et al (2004)³²</td>
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Abbreviations: GRADE = Grading of Recommendations Assessment, Development and Evaluation; ISMAR = Innsbruck Sensorimotor Activator and Regulator.
same positive direction, thereby constituting the basis for a limited recommendation.

Studies With Part of Function as an Outcome. Eleven studies with a total of 321 participants evaluated the effect on motor function in muscles or muscle groups involved in swallowing rather than direct effects on swallowing. The oral sensory-motor stimulation was performed with or without specific therapy tools. The focus for the intervention could be to affect the swallowing pattern, the resting position of lips and tongue, bite force, or occlusion. These studies comprised children and adolescents with different degrees of orofacial dysfunction. The most common diagnosis represented was Down syndrome. Palatal plates were used for the intra-oral sensory-motor treatment of children and adolescents with Down syndrome in 3 studies with moderately strong scientific evidence.15,16,22 All 3 studies showed positive effects on lip and tongue function compared with control subjects. In 3 intervention studies, lip-force training was performed with an oral screen or a FaceFormer (a variant of an oral screen).19,24,29 Four of eight school-aged children with myotonic dystrophy type 1 improved their lip force significantly compared with baseline in a study with moderately strong scientific evidence.24 Forty-five children and adolescents with oral motor dysfunction received oral sensory-motor treatment (unspecified), and 26 of these also used a FaceFormer. All the participants improved their orofacial function, but those who had the additional therapy with a FaceFormer improved more rapidly.30 In an observation study without a control group, 50 children with cerebral palsy, Down syndrome, or intellectual disability used either oral screens or palatal plates.19 Positive effects on orofacial function were reported for both therapy tools. A relationship was found between the time spent on training and the effect of training. The results were stable for 44.6% of the participants 3 months after the intervention ended. Children who participated in a multiple single-study design improved their masticatory function and were able to eat a more varied diet after chewing exercises.28 Seventy-six children and adolescents who had received orthodontic treatment for frontal open bite participated in an observation study with controls.31 Of these, 27 also received myofunctional therapy aimed at correcting the orofacial resting position or swallowing pattern. The risk for relapse after completing the orthodontic treatment decreased in the group of patients who had received myofunctional therapy.

In all included primary studies of interventions for dysphagia, treatment outcomes were based on clinical registrations such as functional feeding skills, food acceptance, and weight. In some studies, electromyography and force measurements were used.17,24,27

Three systematic reviews35-37 and 11 narrative reviews38-48 were identified within the area of dysphagia. In a Cochrane review, Morgan et al16 expressed an urgent need for large-scale randomized studies to evaluate interventions for the treatment of oropharyngeal dysphagia in children and adolescents. Durvasula et al10 pointed to the importance of a multidisciplinary approach in the assessment and treatment of children with dysphagia. Faukles et al42 emphasized the role of the dental team in the treatment of chewing difficulties in patients with Down syndrome. Dusick41 identified 6 different areas that require attention in the management of children with dysphagia: “normalization of posture and positioning, adaptation of foods and feeding equipment, oromotor therapy, feeding therapy, nutritional support and management of associated disorders.” Manikam and Perman43 explained that the causes of pediatric dysphagia are on a continuum between psychosocial and organic factors and believe that, in most cases, they could improve significantly with medical treatment, oral motor training and behavioral therapy. The authors also emphasized that feeding problems are rarely limited to the children but involve the whole family.

Recommendations Relating to Oral Sensory-Motor Interventions for Children and Adults With Dysphagia Secondary to Congenital or Early-Acquired Disabilities. The following recommendations are based on 20 primary intervention studies (Table 1):

- Limited recommendation for intervention with a combination of swallowing therapy and oral sensory-motor stimulation in children with dysphagia secondary to congenital or early-acquired disabilities. The scientific evidence is limited because of a lack of control groups, but congruent, significant positive results constitute the basis of this recommendation.
- Limited recommendation for oral myofunctional therapy after orthodontic treatment of an open bite to prevent a relapse.
- Limited to moderately strong recommendation for oral sensory-motor stimulation (with or without therapy tools) aiming at activating and strengthening the lips, tongue, and chewing muscles in children with orofacial dysfunction.
- Moderately strong recommendation to use ISMAR for the treatment of moderate dysphagia in children with cerebral palsy. The maximum effect of the therapy could be expected within 1 year, and it is probably due to improved jaw stability.
- Moderately strong recommendation to use a palatal plate for the treatment of impaired lip and tongue function in children and adolescents with Down syndrome.
Interventions for Children and Adolescents With Impaired Saliva Control Secondary to Congenital or Early-Acquired Disabilities

Primary Studies. Six primary studies of oral-sensory-motor interventions for the treatment of impaired saliva control were evaluated (Table 2). These studies comprised 312 children and adolescents aged 3 to 17 years. The diagnoses that were represented were orofacial dysfunctions (n = 168),19 Down syndrome (n = 74),19,50 cerebral palsy (n = 56)19,51,52 and intellectual disability (n = 14). In all studies, treatment with an intra-oral appliance, most often a palatal plate, was used. A palatal plate, in isolation or together with other types of oral sensory-motor stimulation, was used for the treatment of impaired saliva control in 5 intervention studies.19,49,50,52,53 They all reported that the intervention resulted in improved saliva control. In an observation study with a control group comprising 68 children with Down syndrome, a clear improvement in saliva control was found compared with the situation before therapy.20 The treatment of impaired saliva control in children and adolescents with cerebral palsy using a palatal plate was evaluated in 2 observation studies without control subjects.19,51 Both studies revealed a positive effect. Improved saliva control was also found in studies including children and adolescents with other disabilities, such as intellectual disability or orofacial dysfunction, but these studies offer insufficient scientific evidence because of the study design.49,51 Treatment with ISMAR had a good effect on 6 children and adolescents with cerebral palsy who participated in an observation study without control subjects. The study had a significant dropout (n = 12), and 1 of the authors is the creator of ISMAR therapy.52 The outcome results in the included studies were based on a clinical evaluation of saliva control.

Systematic and Narrative Reviews. Intervention studies for the treatment of saliva leakage were evaluated in 2 systematic reviews35,54 and 5 narrative reviews.55-59 Arvedson et al35 concluded that evaluations were often complicated because of heterogeneous, poorly described patient groups and that many studies included a combination of interventions. Haberfellner58 and Gisel44 described many years’ experience of treatment with ISMAR and an oral screen. The authors emphasized the importance of improving postural control of the lower jaw, hyoid bone, and tongue and stimulating movements of the lips and tongue to improve salivary control. Crysdale et al59 reported on the results of treatment in 1,487 patients with impaired saliva control. Oral motor therapy was presented as the primary intervention but was often combined with medication or surgery to decrease saliva production.

Recommendations Regarding Oral Sensory-Motor Interventions for Children and Adolescents With Impaired Saliva Control Secondary to Congenital or Early-Acquired Disabilities. The following recommendations are based on 6 primary intervention studies (Table 2):

Moderately strong recommendation to use a palatal plate for the treatment of impaired saliva control in children and adolescents with Down syndrome, cerebral palsy, and/or orofacial dysfunction. Good agreement between available studies strengthens the evidence base.

No recommendation is given regarding the treatment of impaired saliva control with ISMAR, because of insufficient scientific evidence.

Discussion

We evaluated the scientific evidence relating to oral sensory-motor interventions for children and adolescents with dysphagia or impaired saliva control secondary to congenital or early-acquired disabilities. On the basis of the grading of the intervention studies that were included, no strong recommendations could be made. Many of the intervention studies lacked control groups and had other serious limitations relating to study design. The systematic reviews confirmed the insufficient scientific basis for strong recommendations.35-37,54 The literature search ended in February 2016. To our knowledge, no systematic reviews of oral sensory-motor interventions for this patient group were published between March 2016 and August 2018.

Nearly all the identified studies reported good results. However, the study design was often insufficient, and study groups were small. Only a few studies evaluated the effect of oral sensory-motor treatment on saliva leakage, and all these studies included the use of palatal plates or other intra-oral appliances.

In the clinical setting, oral sensory-motor interventions for children with dysphagia or impaired saliva control are often part of a more extensive therapy program involving different team members, such as a physician, a nurse, a physiotherapist, an occupational therapist, a dietician, and a dentist. The management also includes support and practical advice for parents and personnel. The care offered by different team members and cooperation among these providers are often prerequisites for successful interventions.50 The combined and often necessary support of many professionals and the implementation of extensive therapy programs are challenging for clinical research. Other challenges are small, heterogenous patient groups and the ethical aspects of randomized controlled trials.

Each clinician and each clinic can contribute to the common evidence base through the systematic collection and reporting of therapy results. As a professional, the clinician
is expected to learn about each patient’s unique situation and expectations. A detailed knowledge of available interventions and specific knowledge relating to the patient should be integrated and evaluated before any recommendation on clinical management can be presented. The recommendations made from the gradings performed in the present review offer a comprehensive view of a variety of methods available for oral sensory-motor interventions. Individual clinicians will have to determine the applicability of the recommendations to individual patients.61

Conclusion
There is an urgent need for high-quality studies that could serve as the basis for strong recommendations relating to oral sensory-motor interventions for children with dysphagia and impaired saliva control.

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ORCID iD
Lotta Sjögreen https://orcid.org/0000-0001-9551-8686

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