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Pediatric Dysphagia Rehabilitation: Considering the Evidence to Support Common Strategies

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Abstract

Dysphagia in pediatric populations can result in multiple adverse health outcomes. Therefore, childhood swallowing difficulties must be diagnosed accurately and managed appropriately. Effective therapy treatment requires careful consideration of available rehabilitative intervention techniques, and application of strategies that are best suited to rehabilitate the phase(s) of swallowing affected. In pediatric populations, most literature to date has focused on rehabilitative strategies targeting the oral phase of swallowing, often referred to by labels including: oral motor exercises, oral motor interventions, and oral sensori-motor interventions. This article reviews the empirical evidence to support the use of rehabilitative interventions for infants and children with dysphagia, and offers a framework for pediatric clinicians to determine the most appropriate therapeutic targets.

Dysphagia is broadly defined as any disruption to the swallow sequence that results in compromise to the safety, efficiency, or adequacy of fluid and/or nutritional intake (Dodrill & Gosa, 2015). The swallowing process is comprised of three phases: oral, pharyngeal, and esophageal phases. Speech-language pathologists (SLPs) specialize in the management of oral and pharyngeal phase swallowing difficulties.

Approximately one percent of infants and children in the general population will experience oral-pharyngeal dysphagia (OPD; Bhattacharyya, 2015), and OPD may affect up to 80% of infants and children in some clinical populations (Dodrill & Gosa, 2015; Lefton-Greif & Arvedson, 2007). Populations at particular risk include those affected by prematurity (infants born before 37 weeks gestation), respiratory and cardiac disorders, gastrointestinal disorders, and neurological disorders (Dodrill & Gosa, 2015; Lefton-Greif & Arvedson, 2007). OPD can result in adverse sequelae for infants and children, including dehydration, malnutrition, growth faltering, and respiratory complications (Tutor & Gosa, 2012). Due to potential for serious, and sometimes life-threatening, consequences, OPD in pediatric populations must be diagnosed accurately and managed effectively. Any management plan should begin with a thorough assessment to differentiate the phase(s) of the swallow where impairment is occurring, and to allow accurate diagnosis that differentiates the symptoms (i.e., laryngeal penetration) from the physiologic cause of those symptoms (i.e., ineffective laryngeal closure), to facilitate the SLP developing targeted intervention.

In many pediatric populations, the overall goal of OPD therapy management plans is to facilitate the patient achieving safe and adequate oral intake (where possible). This may

necessitate the use of compensatory and/or rehabilitative strategies. *Compensatory interventions* are those strategies that alter the food/fluid bolus or the environment to assist in safe oral intake. Compensatory strategies do not require active participation by the patient. Examples of compensatory strategies include modification of fluid or food texture, specialized feeding equipment, positional changes, and specialized feeding strategies, such as external pacing (Dodrill & Gosa, 2015). *Rehabilitative interventions* are those that are designed to alter the swallowing physiology. The aim of rehabilitative strategies is to repair the damaged swallowing function so that the patient can swallow without the ongoing need for additional compensatory strategies. Due to the physical and cognitive immaturity of infants and children, many of the rehabilitative strategies for OPD pioneered in adult populations are not able to be implemented with the pediatric population.

Oral-Phase OPD Rehabilitative Interventions

Rehabilitative interventions aimed at the oral-phase of swallowing are commonly referred to as oral motor exercises (OME), oral motor interventions (OMI), or oral sensorimotor (OSM) interventions. These may include active motor exercises, passive motor exercises, and sensory motor activities (see Table 1 below for examples) (Arvedson, Clark, Lazarus, Schooling, & Frymark, 2010a).

Table 1. Examples of Oral-Phase Rehabilitative Interventions.

Category	**Definition**	**Examples**
Active motor exercises	Movement of a specific body part (e.g., lips) initiated by voluntary contraction and relaxation of the muscles controlling that body part Used to improve strength and/or endurance	• Stretching • Strength training
Passive motor exercises	Movement initiated to a specific body part (e.g., cheeks) by another individual or tool; Used to increase sensory input or enhance flexibility in joints (e.g., tempro-mandibular joint)	• Massage • Passive range of motion
Sensory-motor activities	Application of a sensory input (e.g., tactile, thermal) or other sensory variable (e.g., electrical current) to a specific body part (e.g., tongue) to modulate sensory registration and/or to improve motor response to facilitate safer/more efficient swallowing	• Neuromuscular electrical stimulation (NMES) • Use of vibratory oral tools (e.g. tooth brushes)

There are many different types of oral-phase rehabilitative interventions used by pediatric SLPs on a day-to-day basis and featured in clinical manuals on pediatric dysphagia. However, the evidence supporting these practices is often limited by issues such as small sample sizes, varied populations, and varied intervention techniques (including varied dose, frequency, and length of treatment). The following section of this paper will review the use of common strategies, and outline the evidence to support their use in various pediatric populations with OPD.

Use of Oral-Phase OPD Rehabilitative Interventions in Preterm Infants

Due to immaturity of major body systems involved in feeding (e.g., brain, lungs, and gut), as well as exposure to noxious sensory stimulation to the oralpharyngeal region (e.g., as caused by intubation and gavage feeding tubes), preterm infants are at risk of feeding difficulties affecting sucking (oral-phase of swallowing) and suck-swallow-breath coordination (pharyngeal-phase of swallowing). However, most therapy interventions described in the literature with this population to date have focused on oral-phase function. Many of the interventions described

utilize passive motor exercises and sensory motor activities involving the oral cavity and perioral region. The aim of these exercises is generally to encourage tolerance of oral-facial stimulation and to promote non-nutritive sucking (NNS) before the infant is developmentally or medically ready to initiate nutritive sucking and oral feeding. There is a large body of literature reporting on the effects of oral-phase rehabilitative interventions in preterm populations, including three systematic reviews.

In 2010, Arvedson and colleagues published a systematic review on the effects of various oral-phase rehabilitative interventions on the swallowing and feeding skills of infants born prematurely. This review focused on interventions involving opportunities for NNS, oral/perioral sensory stimulation, and combinations of these techniques. NNS can be facilitated with gloved finger and/or pacifier, and the aim of these exercises is to allow sucking practice. Oral/perioral stimulation programs involve the use of tactile stimulation to the face and oral cavity with the aim of decreasing oral hypersensitivity and improving tone and range of movement of oral structures. Arvedson et al. (2010b) identified twelve studies that met their criteria. Based on a review of these articles, they concluded that while there is some promising preliminary evidence that oral-phase rehabilitative interventions may positively influence the feeding/swallowing skills of preterm infants, limitations to the existing evidence base (variations in types of interventions used, length of intervention provided, and variations in the diagnoses and maturation of the infants to whom the interventions were applied, in combination with reported mixed results) should encourage clinicians to carefully consider the potential benefits vs. costs and risks of utilizing these types of interventions in such medically fragile populations (Arvedson et al., 2010b).

Since the publication of the Arvedson et al. (2010b) review there have been multiple papers published that continue to examine the use of oral-phase rehabilitative interventions for preterm populations. Several papers report on the use of a specially developed synthetically patterned (mechanical) orocutaneous input tool that targets NNS (Barlow, Jegatheesan, Weiss, et al., 2014; Barlow, Lee, Wang, et al., 2014; Song et al., 2014). This literature suggests that the synthetically patterned oral input has a positive impact on NNS in preterm infants (in terms of bursts per minute, NNS events per minute, and total compressions per minute; Barlow, Lee, et al., 2014). In addition, this literature suggests that the synthetically patterned oral input modulates the electroencephalography (EEG) output in preterm infants, and has the potential for to influence thalamacortical and corticocortical development (Barlow, Jegatheesan, Weiss, et al., 2014; Song et al., 2014). However, this research has not clearly established the functional benefit of this type of oral stimulation on nutritive sucking or swallow safety (Barlow, Jegatheesan, Weiss, et al., 2014; Song et al., 2014).

Six recent papers have investigated the effects of various combinations of oral-phase rehabilitative interventions in relation to a variety of feeding and swallowing outcomes in preterm infant populations, including the following: proficiency of feeding (volume of intake during the first five minutes of feeding), behavioral state, time to transition to full oral feeding, length of hospital stay, breastfeeding rates, and weight gain (Bache, Pizon, Jacobs, Vaillant, & Lecomte, 2014; Coker-Bolt, Jarrard, Woodard, & Merrill, 2013; Hwang et al., 2010; Lessen, 2011; Liu et al., 2013; Younesian, Yadegari, & Soleimani, 2015; Zhang et al., 2014). Results are summarized in the table below (Table 2).

Table 2. Summary of Reported Effects of Oral-Phase Rehabilitative Interventions in Preterm Populations From Recent (published after 2009) Literature.

Outcome of Interest	**Reported Effect**
Proficiency of feeding (volume of intake during the first five minutes of feeding)	• Infants receiving pre-feeding oral stimulation program achieved better proficiency of oral feeding compared to controls (Hwang et al., 2010) • In a comparison of NNS, oral stimulation (OS), a combination of NNS and OS, and controls, oral feeding proficiency was greater in the combination NNS and OS group, as compared to all other groups (Hwang et al., 2010)**
Behavioral state	• A higher percentage of infants receiving pre-feeding oral stimulation program transitioned to a drowsy or quiet alert state from sleep or restless state before feeding, as compared to controls (Hwang et al., 2010)
Time to transition to full oral feeding	• Oral sensory motor stimulation program applied to preterm infants between 30–32 weeks of gestational age resulted in earlier transition to full oral feeding (Younesian et al., 2015) *Note: GA at full oral feeding 32.8/40 (intervention, n=10) vs 34.5/40 weeks GA (control, n=10). Mean GA at birth = 30–31/40 weeks GA.* • Premature infants receiving the Premature Infant Oral Motor Intervention (PIOMI) program once per day transitioned to full oral feeding sooner (Lessen, 2011). *Note: GA at full oral feeding 34.1/40 (intervention, n=10) vs 34.5/40 weeks GA (control, n=9). Mean GA at birth = 28/40 weeks GA.* • NNS, OS, and a combination of NNS and OS resulted in reduced time to transition to full oral feeding (Zhang et al., 2014)** *Note. GA at full oral feeding 34.8/40 weeks GA (NNS, n=29), 35.1/40 (OS, n=27), 34.6/40 (NNS+OS, n=29) vs 35.4/40 (control, n=27). Mean GA at birth=30–31/40 weeks GA.* • Early oral stimulation program administered before beginning oral feedings resulted in no difference in the time needed to transition to full oral feeding (Bache et al., 2014)** *Note: GA at full oral feeding 36.4/40 (intervention, n=40) vs. 36.3/40 weeks GA (control, n=46). Mean GA at birth=31/40 weeks GA.*
Length of hospital stay	• Oral motor management program administered to preterm infants resulted in shorter duration of hospital stay (Liu et al., 2013). *Note. Mean discharge age=36.5/40 weeks GA (intervention) vs. 37.7/40 weeks GA (control)* • Oral sensory motor stimulation program applied to preterm infants between 30–32 weeks GA resulted in reduced length of hospital stay as compared to infants in a control group (Younesian et al., 2015). *Note. No data presented on GA at discharge; Mean discharge age day of life=27.9 (intervention) vs. 38.8 (control)* • Premature infants receiving the PIOMI program were discharged sooner than infants in the control group (Lessen, 2011). *Note. No data presented on GA at discharge; outliers removed from intervention group* • NNS, OS, and a combination of NNS and OS resulted in no difference in length of hospital stay as compared across all three intervention groups and a control group (Zhang et al., 2014)** • Early oral stimulation program administered before beginning oral feedings resulted in no difference in length of hospital stay (Bache et al., 2014)**

(continued)

Breastfeeding rates	• Early oral stimulation program administered before beginning oral feedings resulted in a higher percentage of breastfeeding rates upon discharge (Bache et al., 2014)** *Note: Percentage discharged on partial or total breastfeeding= 70% (intervention) vs. 46% (control)*
Weight gain	• Oral sensory motor stimulation program applied to preterm infants between 30–32 weeks GA resulted in no difference in weight gain between the experimental and control group (Younesian, Yadegari, & Soleimani, 2015) NNS, OS, and a combination of NNS and OS resulted in no difference in weight gain as compared across all three intervention groups and a control group (Zhang et al., 2014)**

Note. **randomized clinical trial.

In 2015, two additional systematic reviews were published on the effects of oral-phase rehabilitative interventions on preterm infants' oral feeding performance. The first, published in early 2015, summarized 29 publications (consisting of ~45% clinical trials; Lima, Cortes, Bouzada, & Friche, 2015). The second, published in late 2015, reviewed 11 randomized controlled trials (Tian et al., 2015). Both concluded that oral-phase rehabilitative interventions in preterm infants may shorten the transition time to full oral feeding. The impact of these interventions on other clinically relevant outcomes (e.g., swallow safety, physiological stability, and length of stay) remains unclear.

It should also be considered that, as clinicians are applying oral-phase rehabilitative interventions programs focused on improving feeding and swallowing abilities, they are also, perhaps unintentionally, providing concomitant sensory stimulation to other developing sensory systems including the auditory, vestibular, visual, and olfactory systems. Feeding and swallowing are perhaps the most sensory rich activities that infants participate in, and the experience is not limited solely to oral stimulation, as the young infant is completely dependent on the feeder for positional support during the feeding. Unable to completely control for this additional stimulation, and recognizing the importance of the other sensory systems' potential influence on feeding and swallowing outcomes, recent papers have attempted to acknowledge the other types of sensory stimulation the premature infant may be receiving during the focused oral-phase rehabilitative interventions, and quantify the impact of the complete sensory stimulation being provided. In a series of publications, Fucile and colleagues randomized a sample of 75 preterm infants (gestational age [GA] at birth <32/40 weeks) to one of three interventions: oral (n=19), tactile/kinesthetic (n=18), and combined oral and tactile/kinesthetic intervention programs (n=18) vs. control (n=20; Fucile & Gisel, 2010; Fucile, Gisel, McFarland, & Lau, 2011; Fucile, McFarland, Gisel, & Lau, 2012). They reported improved transition to full oral feeding in all the intervention groups (mean GA at full oral feeding = 35.9/40 oral, 35.4/40 tactile/kinesthetic, 34.7/40 oral and tactile/kinesthetic, vs. 36.2/40 control; Fucile et al., 2011; Fucile et al., 2012). Medoff-Cooper and colleagues (2015) also investigated the effects of a multisensory intervention (in this case, the auditory, tactile, visual, and vestibular [ATVV] intervention) on sucking organization. A sample of 183 preterm infants (GA at birth 29–34/40 weeks GA) were randomized to the intervention (n= 90) vs. control (n=93). Those receiving the ATVV intervention were reported to demonstrate improved sucking frequency and pressure relative to the control group over a 14 day period. However, the GA at full oral feeding is not reported, so it is not possible to evaluate if this intervention ultimately reduced transition time to full oral feeding (Medoff-Cooper et al., 2015).

Use of Oral-Phase OPD Rehabilitative Interventions in Other Infants and Children

Arvedson and colleagues (2010a) published a systematic review on literature published before September 2007 that sought to determine the effects of oral-phase rehabilitative interventions on swallowing outcomes (functional feeding ability, drooling, swallowing physiology, and pulmonary health) in children other than preterm infant populations. The authors identified 16 studies that met their criteria (of note, none of the studies reported on pulmonary health outcomes). The

identified studies reported mixed intervention results across children of various ages with varying diagnoses (including cerebral palsy, Trisomy 21, and mental retardation). The studies were of inconsistent methodological quality, and reported on multiple different types of intervention techniques. The authors concluded that there is insufficient evidence for, or against, the use of oral-phase rehabilitative interventions (active motor exercises, passive motor exercises, and/or sensory-motor activities) for the improvement of swallowing and/or feeding skills in older children (Arvedson et al., 2010a).

More recently, a study was published describing the use of an oral motor stimulation program for infants born with congenital heart disease (univentricle anatomy; Coker-Bolt et al., 2013). Infants receiving the intervention group (n=18) were compared to a historical control group (n=10). The authors report that those receiving the intervention reached full oral feeding two days faster than their control counterparts (6.3 vs. 8.3 days), and discharged seven days sooner (28.6 vs. 35.3 days), though it is unclear if there were other confounding variables that impacted on results.

Pharyngeal-Phase OPD Rehabilitative Interventions

As mentioned previously, many of the rehabilitative strategies for OPD pioneered in adult populations are not able to be implemented with the pediatric population, due to the physical and cognitive immaturity of infants and children. This is particularly the case for many active pharyngeal-phase interventions, which require an awareness of the swallow process to implement learned strategies (e.g., supra-supraglottic swallow). There is one peer-reviewed study that focuses on the application of a passive pharyngeal-phase intervention, specifically, neuro-muscular electrical stimulation (NMES) of anterior neck muscles, in pediatric patients with OPD (Christiaanse et al., 2011). In this study, the authors compared swallow function in a group that received NMES in addition to diet modification and oral feeding practice (n=46) with a historical control group who received diet modification and oral feeding practice, but not NMES (n=47). The authors concluded that, in a heterogeneous group of children with OPD, NMES did not improve swallow function.

There have been two systematic review papers that have focused on oral-phase and pharyngeal-phase rehabilitation exercises in specific groups of children with OPD. Morgan, Dodrill, and Ward (2012) conducted a Cochrane review to investigate the evidence to support the use of oral/pharyngeal rehabilitation exercises in the management of OPD in children with neurological impairment (i.e., acquired brain injury, genetic syndromes, and degenerative conditions). The authors identified only three studies that met their inclusion criteria, and their analysis concluded that there was currently insufficient empirical evidence to support or refute the use of any specific oral/pharyngeal rehabilitation exercises for children with neurological impairment (Morgan et al., 2012). Harding and Cockerill (2015) reviewed the effectiveness of oral/pharyngeal rehabilitation exercises in the management of OPD in children with learning disabilities. Their analysis concluded that there was a lack of evidence to support or refute the use of oral/pharyngeal rehabilitation exercises in this population also (Harding & Cockerill, 2015).

Conclusions

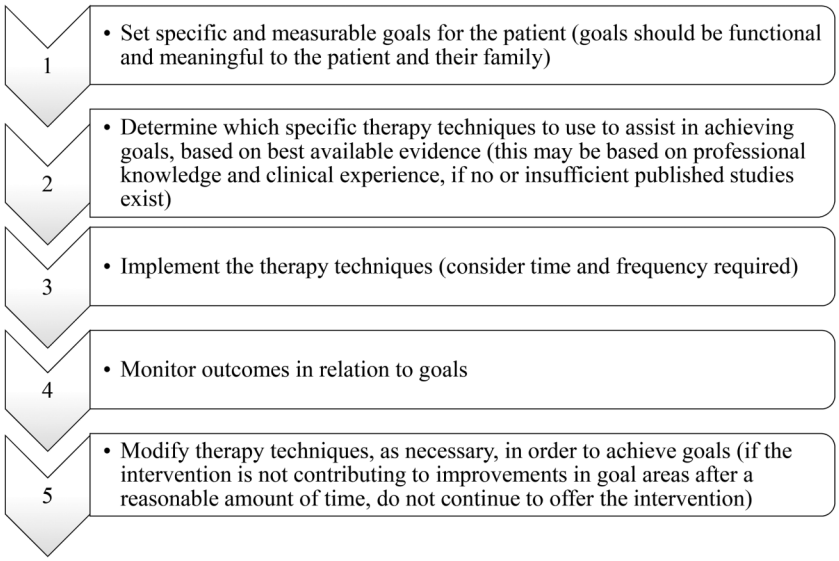
There are many different types of rehabilitative interventions targeting the oral-phase and pharyngeal-phase of swallowing utilized by pediatric SLPs. However, it must be acknowledged that the current evidence supporting these interventions is limited, and affected by issues such as small sample sizes, varied populations, and varied intervention techniques. Knowing this, pediatric SLPs may feel at a loss for what to do to assist their patients with OPD.

It is important to remember that any feeding therapy management plan should begin with a thorough assessment to differentiate the phase(s) of the swallow where impairment is occurring, and to elucidate both the symptoms and the physiologic cause of those symptoms, in order to facilitate appropriate and targeted intervention. In order to facilitate the pediatric patient achieving

safe and adequate oral intake, both use of compensatory and rehabilitative strategies may be necessary, at least in the short-term. In cases where the patient/family are committed to working towards achieving full feeding function, and it has been assessed that this is an achievable long-term goal, the focus of therapy intervention should be on improving sensory motor activity and reducing the need for compensatory strategies.

In the absence of a strong evidence -base for many OPD therapy techniques, SLPs must thoughtfully consider the underlying rationale for any interventions that are implemented with patients. In addition, SLPs must apply the principles of scientific therapy practice, as outlined in Figure 1. Applying these principles in pediatric OPD practice ensures that clinicians are carefully considering the results of accurate diagnostic assessments, and choosing therapeutic targets that are specific and individualized to the patient's need. Clinicians should not apply standard "recipes" without considering patient variables, or apply interventions that are not targeted at the area of impairment.

Figure 1. Principles of Scientific Therapy Practice.



Source. Reprinted with permission from Dodrill, 2015.

Pediatric SLPs are uniquely qualified to assist infants and children with OPD due to their combined knowledge of feeding development, swallowing anatomy, physiology, and neural control, and their professional commitment to evidence-based practice. In the best interest of patient care and professional standards, clinicians and researchers must commit to documenting and sharing the results of their clinical practice. In order to allow appropriate evaluation and critique of intervention techniques, well-designed clinical trials are required to address existing gaps in the literature.

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