

## Research Article

# Oral Sensory-Motor Intervention for Children and Adolescents (3-18 Years) with Developmental or Early Acquired Speech Disorders – A Review of the Literature 2000-2017

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**Abstract**

**Objectives:** The aim of this study was to investigate the level of scientific evidence for different sensory-motor interventions in children and adolescents with motor speech disorders. A final aim was to identify key motor learning principles in these methods.

**Methods:** A review of the literature published 2000-2017, including sensory-motor intervention for children and adolescents (3-18 years) with congenital, developmental or early acquired disabilities affecting speech, was performed. The literature search included the databases PubMed, CINAHL, Medline, Speech BITE, OVID, ERIC, Cochrane and Google Scholar. Identified primary studies were evaluated using Grading of Recommendations, Assessment, Development and Evaluation.

**Results:** Forty-four studies, reporting results following 16 sensory-motor intervention methods for different motor speech disorders, fulfilled inclusion criteria. Of these, three were randomized controlled trials. Scientific evidence varied between strong and insufficient. Rapid Syllable Transition Treatment was the only method rated as high scientific evidence for children with childhood apraxia of speech. Several methods reached medium high scientific evidence with studies reporting improved speech accuracy, loudness or voice quality. The most common motor learning principle was high frequency interventions with  $\geq 20$  sessions/week. Children with syndromes had longer reported treatment periods.

**Conclusions:** Rapid Syllable Transition Treatment is recommended for children with childhood apraxia of speech. In several patient groups evidence is low or lacking consequently there is a need for high quality intervention studies to guide clinicians.

**ABBREVIATIONS**

CAS: Childhood Apraxia of Speech; SSD: Speech Sound Disorders; MSD: Motor Speech Disorders; EBP: Evidence-Based Practice; RCT: Randomized Controlled Trial; GRADE: The Grading of Recommendations Assessment Development and Evaluation; ReST: Rapid Syllable Transition Treatment; NDP3: Nuffield Dyspraxia Program3; DTTC: Dynamic Temporal and Tactile Cueing; PROMPT: Prompts for Restructuring Oral Muscular Phonetic Targets; MSTP: Motor speech treatment protocol; MSI:

Motor Speech Impairment/involvement; EPG: Electro Palato Graphy; DS: Down syndrome; CP: Cerebral Palsy; LSVT LOUD: Lee Silverman Voice Treatment – LOUD; TBI: Traumatic Brain Injury; NSOM: Non-Speech Oral Motor Treatment; PPT: Phonetic Placement Therapy; sEMG: surface Electro-MyoGraphy; SLP/SLT: Speech and Language Pathologist/Therapist

**INTRODUCTION**

Speech disorders may hamper several aspects of communication, participation and quality of life [1]. During

childhood, speech disorders are among the most common developmental disorders affecting between 10-25% depending on age, type and severity of the disorder [2-5].

The two sensory-motor based speech disorders affecting children and teenagers are childhood apraxia of speech (CAS) and dysarthria. CAS is regarded as primarily affecting speech motor planning and programming [6]. Dysarthria is a speech disorder including difficulties controlling the muscles needed for speaking [7]. Children with dysarthria often have impaired control of articulators and shallow, irregular breathing which creates difficulties generating sufficient breath to support speech. Also speech sound disorders (SSD) may have sensory-motor involvement.

Interventions for motor speech disorders (MSD), aim to establish or reestablish appropriate speech movements. The most fundamental principle has been suggested to be amount of practice [8]. However, several other aspects may also influence treatment outcome. A central question in motor speech interventions has been whether principles based on interventions for other motor functions also apply to the treatment of MSD. Furthermore, an impaired sensory motor system may not respond to the same principles as a healthy one [9]. In this tutorial, Maas and coworkers [9] conclude that "... various principles may interact with each other and differentially affect diverse aspects of movements" [9] Table 1 and 2, p 282). Accordingly, the optimal intervention and design for different patient groups with MSD is debated.

Evidence-based practice (EBP) is a key factor in providing patients with the best services. The need to arrive at a clinical decision has promoted the development of EBP. Cochrane reviews, other systematic reviews, meta-analyses and narrative reviews play an important role in building such evidence and aiding the clinician. However, to full-fill quality criteria for Cochrane reviews the study needs to be a randomized controlled trial (RCT). Within the area of speech and language pathology RCT studies are currently scarce.

In an attempt to provide easily accessible reviews of currently available best evidence the association of Swedish habilitation leaders in 2001 initiated a review process of the literature primarily intended for professionals working with children within the habilitation services. All reports are written in Swedish and freely available on their web-site. This study was part of such a report [10]. The original project also included interventions for dysphagia and impaired saliva control [11].

The aim was to evaluate and describe currently available best evidence for sensory-motor interventions in children and adolescents 3 to 18 years old with developmental or early acquired speech disorders. Based on the review, we also aimed at providing recommendations for clinicians. A final aim was to identify motor learning principles for recommended intervention methods.

## METHODS

Inclusion criteria were: (a) articles published in scientific peer-reviewed journals between 2000-01-01 and 2017-02-29, (b) including children three to 18 years old, (c) diagnosed with,

developmental or an early acquired speech disorder (dysarthria, CAS or SSD), (d) reporting effects of sensory motor intervention, (e) written in English, Swedish or German.

Included study designs were RCT studies, multiple and single-subject/case studies, cohort studies, case series and observational studies. Single-subject studies were included since early evidence for a method can be reported using this method. Expert opinions or reviews were not included.

A literature search was performed using the databases PubMed, CINAHL, Medline, Speech BITE, OVID, ERIC, Cochrane and Google Scholar. Papers that fulfilled inclusion criteria were included in the review.

The Grading of Recommendations Assessment, Development and Evaluation, GRADE, was used to assess the quality of included studies [12-14]. GRADE uses a four steps interval scale ranging from high (4), to moderate (3), limited (2) or insufficient quality (1). The highest rated study designs are RCT studies, meta-analyses and systematic reviews (GRADE 4) [14]. Controlled trials are rated as moderate quality (GRADE 3). Observation studies without controls and studies with a multiple single-subject design (MSSD) are considered to be of limited quality (GRADE 2) and case reports are rated as insufficient quality (GRADE 1). Depending on factors increasing or decreasing the quality of the study, the initial GRADE value can be increased or decreased with up to two points.

The recommendations for an intervention method and 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 high quality and no or few diminishing factors resulting in a strong recommendation.
- *Medium high scientific support:* Methods with high or medium high quality with diminishing factors in the overall assessment of the assessed studies resulting in a moderately strong recommendation.
- *Limited scientific support:* Method with high or medium high quality and substantial diminishing factors in the overall assessment of the assessed studies resulting in a limited recommendation.
- *Insufficient scientific support:* Methods where the number of studies was few or of low quality or available studies reported conflicting results. No recommendation could be made.

Finally, motor learning principles for the recommended methods were evaluated based on principles suggested by Maas and coworkers (2009) [9]. The following motor learning principles were included 1) number of treatment sessions/week, 2) total number of treatment sessions, 3) length/session, 4) dose or number of repetitions/session, 5) treatment design (randomized or blocked practice), 6) simple or complex targets, 7) who conducted the treatment, 8) how was feedback provided, 9) were there any generalization effects and 10) were there any reports on stability of treatment results according to follow-up.

**Table 1:** Studies reporting results for children and adolescents with apraxia of speech/CAS or SSD. Authors, intervention methods, study design, age and number of included participants, type of disorder, additional disabilities if reported and GRADE is presented.

| Reference                                   | Intervention                                                        | Participants |            | Diagnosis/<br>Dysfunction       | GRADE |
|---------------------------------------------|---------------------------------------------------------------------|--------------|------------|---------------------------------|-------|
|                                             |                                                                     | Sample size  | Age, y     |                                 |       |
| Murray et al., 2015 <sup>16</sup>           | ReST&NDP3                                                           | 26           | 4-12       | Mild-severe CAS                 | 4     |
| McCabe et al., 2014 <sup>18</sup>           | ReST                                                                | 4            | 5:5-8:6    | CAS                             | 2     |
| Thomas et al., 2014 <sup>17</sup>           | ReST                                                                | 4            | 4:8-8:0    | CAS                             | 2     |
| Ballard et al., 2010 <sup>15</sup>          | Syllable repetition ( $\approx$ ReST)                               | 3            | 7-10       | CAS                             | 2     |
| Baas et al., 2008 <sup>22</sup>             | DTTC                                                                | 1            | 12:8       | CAS & CHARGE                    | 2     |
| Strand et al., 2006 <sup>21</sup>           | DTTC                                                                | 4            | 5:5-6:1    | CAS                             | 2     |
| Maas, Farinella, 2012 <sup>23</sup>         | DTTC                                                                | 4            | 5:0-7:9    | CAS                             | 2     |
| Maas et al., 2012 <sup>24</sup>             | DTTC                                                                | 4            | 5:4-8:4    | CAS                             | 2     |
| Skelton, Hagopian, 2014 <sup>25</sup>       | DTTC                                                                | 3            | 4-6        | CAS                             | 2     |
| Edeal et al. 2011 <sup>19</sup>             | DTTC                                                                | 2            | 3:4, 6:2   | CAS                             | 1     |
| Strand, Debortine, 2000 <sup>20</sup>       | Integrated stimulation ( $\approx$ DTTC)                            | 1            | 5:9        | CAS                             | 1     |
| Square et al., 2014 <sup>27</sup>           | PROMPT                                                              | 5            | 4:0-4:9    | SSD; SSD with MSI               | 2     |
| Yu et al., 2014 <sup>28</sup>               | PROMPT                                                              | 6            | 5:1 (mean) | SSD                             | 3     |
| Dale, Hayden, 2013 <sup>29</sup>            | PROMPT                                                              | 4            | 3:6-4:8    | CAS                             | 2     |
| Namasivayam et al 2013 <sup>26</sup>        | PROMPT                                                              | 12           | 3:11-6:7   | CAS                             | 2     |
| Namasivayam et al 2015 <sup>31</sup>        | MSTP                                                                | 5            | 3:2-3:5    | SSD with MSI                    | 2     |
| Namasivayam et al 2015 <sup>30</sup>        | MSTP                                                                | 37           | 2:8-4:6    | CAS                             | 3     |
| Martikainen, Korpilahti, 2011 <sup>35</sup> | Melodic Intonation Therapy; Touch-Cue Method                        | 1            | 4:7        | CAS                             | 2     |
| Iuzzini, Forrest, 2010 <sup>34</sup>        | Stimulability training protocol; modified core vocabulary treatment | 4            | 3:7-6:10   | CAS                             | 2     |
| Forrest, Iuzzini 2008 <sup>36</sup>         | NSOMs; production training                                          | 10           | 3:6-6:3    | SSD, phonology/<br>articulation | 1     |
| Koskimieset al., 2011 <sup>32</sup>         | Palatal plate                                                       | 168          | 4-14       | SSD; SSD with MSI               | 1     |
| McAllister 2003 <sup>33</sup>               | Palatal plate                                                       | 38           | 4-9        | SSD with MSI                    | 1     |
| Lundeborg, McAllister 2007 <sup>37</sup>    | Intra-oral sensory stimulation; EPG                                 | 1            | 5:0        | CAS                             | 1     |

**Abbreviations:** GRADE: Grading of Recommendations Assessment Development and Evaluation; ReST: Rapid Syllable Transition Treatment; NDP3: Nuffield Dyspraxia Program3; CAS: Childhood Apraxia of Speech; DTTC: Dynamic Temporal and Tactile Cueing; PROMPT: Prompts for Restructuring Oral Muscular Phonetic Targets; SSD: Speech Sound Disorders; MSI: Motor Speech Involvement; MSTP: Motor speech treatment protocol; NSOMs: Non-speech Oral Motor Treatment; EPG: Electro Palato Graphy

**Table 2:** Studies reporting treatment outcomes for children and adolescents with Down syndrome. Authors, intervention methods, study design, age, number of included participants, and GRADE is presented.

| Reference                            | Intervention                    | Participants<br>Sample size | Age, y          | GRADE |
|--------------------------------------|---------------------------------|-----------------------------|-----------------|-------|
| Carlstedt et al., 2001 <sup>38</sup> | Palatal plate; no palatal plate | 20                          | 5:6 y $\pm$ 1.5 | 3     |
| Carlstedt et al., 2003 <sup>39</sup> | Palatal plate; no palatal plate | 20 (same as above)          | 9:6 y $\pm$ 1.5 | 3     |
| Bäckman et al., 2007 <sup>40</sup>   | Palatal plate; no palatal plate | 68                          | 4 y $\pm$ 6 m   | 2     |
| Cleland et al., 2009 <sup>41</sup>   | EPG                             | 6                           | 10:1 – 18:9     | 2     |
| Wood et al., 2009 <sup>42</sup>      | EPG                             | 2                           | 11:7; 14:11     | 1     |

**Abbreviations:** GRADE: Grading of Recommendations Assessment Development and Evaluation; EPG: Electro Palato Graphy

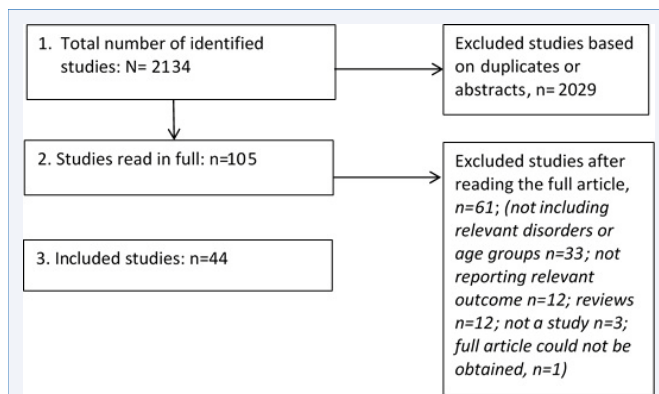
## RESULTS

A total of 2128 studies were identified from the database searches, six studies were added after a manual search resulting in a total of 2134 studies, see Figure 1.

After the deletion of all duplicates and obviously irrelevant studies, the full article from the remaining 105 were read and

categorised according to participants, diagnosis and type of intervention. Only studies that related to an oral sensory-motor intervention including at least one child with dysarthria, CAS or SSD were finally selected for the review. Through this process, the total number of included studies were 44, see Figure 1.

All studies were first graded independently. To investigate interjudge reliability between graders, Cronbach's alfa was



**Figure 1** Search process, exclusions and the final number of included studies based on decided criteria. In the first step doubles were excluded followed by exclusions based on title, key words and abstract including relevant methods and populations. In the second step exclusions were made based on including relevant age groups, reporting results from a sensory motor intervention according to criteria and being a primary study. One article could not be obtained in full leaving the final number of included studies to n=44 as shown in step three. The studies report results following sensory motor intervention for children with SSD (n=6), CAS (n=17) and dysarthria (n=21).c

calculated for 17 randomly selected studies (39%). The selected studies were graded by one other investigator. Interjudge reliability was high with an alpha value of  $\alpha = .984$  indicating excellent agreement.

The study designs represented in included studies were three randomized controlled trials, 31 multiple or single subject design studies, six observational studies, three case series and one retrospective study. A total of sixteen different sensory-motor intervention methods were used in the included studies.

### Interventions for children and adolescents with CAS or SSD

In total, 22 primary studies described effects of sensory-motor interventions for children and adolescents with CAS or SSD, see Table 1. The studies included 347 individuals, age 3:4 to 14 years, 230 children with SSD and 117 with CAS. Thirteen different methods were described and used separately or in combination. Four studies, including a total of 37 children, 4 to 12 years, described effects following interventions using Rapid Syllable Transition Treatment (ReST) [16-18]. All four studies reported positive effects after intervention. ReST and Nuffield Dyspraxia Program3 (NPD3) were compared in a RCT study with 13 children randomized to each method [17]. The authors reported significant treatment effects following both interventions. The long-term follow-up showed stronger effects for ReST with a continued improvement after treatment ended. The GRADE for the ReST studies varied from 4 for the RCT study to 2 for the other studies. The GRADE for NPD3 was 3 since the method was used in only one study with a limited number of participants.

Dynamic Temporal and Tactile Cueing (DTTC), or a predecessor to the method, was used in seven studies including 19 individuals 3:4 to 12:8 years [19-25]. A total of 16 children

improved from the intervention. The GRADE assigned the DTTC studies varied from 1 to 2.

Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT) was used in four studies including 27 individuals between 3:6 and 6:7 years [26-29]. All participating children improved speech and/or speech related functions such as voice onset time after the intervention. In two studies, one of the founders of the method was involved, increasing the risk of bias regarding reported results. The assigned GRADE for the PROMT studies varied between 2 and 3.

Namasivayam and coworkers [30] investigated the effects of the Motor speech treatment protocol (MSTP) in 37 children 2:8 to 4:6 years old with CAS. The number of treatment sessions/w were compared [30]. Significant improvements for articulation and functional communication were reported for the group that received treatment twice/w compared to once/w. Neither intervention intensity yielded a significant change in speech intelligibility at the word or sentence level. Effect sizes according to Cohen's d were larger for most variables following a twice/w intervention. MSTP was also used with five children 3;2 to 3:5 years with severe to profound SSD [31]. Four of the five children demonstrated significant change between baseline and maintenance conditions. Both practiced and non-practiced test-words demonstrated improvement in all participants, indicating generalization of target features to untrained words. Effect size was large for intelligibility at word level.

Koskimies, Pahkala and Myllykangas (2011) [32] and McAllister (2003) [33] investigated effects of palatal plate training in 168 and 38 children respectively, with mild to moderate oral dysfunctions and SSD with motor speech involvement (MSI). The children were between 4 and 14 years old. Results reported by Koskimies and coworkers in their retrospective study were improved speech/articulation in 51% of the participants and improved tongue mobility and lip-closure in 47% and 38% respectively [32]. The study group was heterogeneous and superficially described. Results in the study of 38 children were significantly improved voice quality following the intervention aimed at improved speech and oral motor function [33]. No details regarding speech characteristics were provided. Both studies were assigned GRADE 1.

Three studies reported effects following a combination of treatments. Izzuini and Forrest (2010) [34] investigated the impact of a dual treatment approach that included a stimulability training protocol (STP) and a modified core vocabulary treatment (mCVT) for speech sounds in four children with CAS. Forrest and Izzuini also explored effects of oral motor training or NSOMs compared to traditional production training in ten children with SSD [35]. Effects following Melodic Intonation Therapy and the Touch-Cue Method were compared by Martikainen and Korpilahti (2011) [36] for one child with CAS and Lundeborg and McAllister (2007) [37] investigated effects of a combined treatment of intra oral sensory stimulation and electropalatography (EPG) in one child with CAS. All four studies reported positive effects on speech following intervention. Forrest and Izzuini (2008) reported significantly greater production gains with traditional production treatment compared to NSOMs [35]. Assigned GRADEs varied between 1 and 2 for the combined treatment studies.

Recommendations regarding oral sensory-motor interventions for children and youth with CAS or SSD based on assessed intervention methods are:

- *Medium high to high scientific support* to use ReST for children and adolescents with CAS
- *Medium high scientific support* to use NDP3 for children and adolescents with CAS
- *Medium high scientific support* to use DTTC for children and adolescents with CAS
- *Medium high scientific support* to use PROMPT for children and adolescents with CAS or SSD
- *Medium high scientific support* to use MSTP for children and adolescents with CAS

For other described intervention methods, the scientific support is insufficient.

### Methods for children and youth with dysarthria and Down syndrome

Five studies described effects of interventions for children and adolescents with dysarthria and Down syndrome (DS), Table 2. The studies included 96 individuals between 4 and 18:9 years. Three studies described interventions using palatal plates ad modum Castillo Morales for 56 children 4 to 9:6 years old [37-39]. Two were RCT studies [38-39] with one study also reporting on longitudinal effects after four years of treatment [40]. The RCT studies included the same 20 children with DS, with nine randomized to palatal plate treatment. The observation study included 37 children in the treatment group and two control groups, one with children with DS not treated with palatal plate (n=31) and one with typically developing children (n=36) [40]. All three studies reported positive effects on speech or functions related to speech such as lip-rounding, tongue position or mobility.

Two studies, including a total of 8 individuals between 10 and 18 years, reported effects after interventions using EPG [41-42]. The authors reported significant treatment effects including improved intelligibility, vowel and consonant accuracy and words,

Recommendations regarding oral sensory-motor interventions for children and youth with dysarthria and DS based on assessed intervention studies are:

- *Limited to medium high scientific support* to use palatal plates for children with dysarthria and DS.

For other described intervention methods, the scientific support is insufficient. Reported results following treatment with EPG are promising.

### Methods for children and adolescents with dysarthria due to cerebral palsy or traumatic brain injury

Fifteen studies were identified describing effects of intervention for children with cerebral palsy (CP) with or without other disorders, see Table 3. The studies reported results for 108 individuals between 3 and 18 years old using seven different intervention methods or a combination of methods.

Lee Silverman Voice Treatment – LOUD (LSVT LOUD) was used in four studies including in total 17 children between 3 and 10 years [43-46]. LSVT LOUD targets strength and loudness and the coordination of respiratory, laryngeal, and supralaryngeal systems of speech mechanism. Reported results vary somewhat across participants in the four studies. The number of treated individuals in each study is low but methodology and results are well presented and include both perceptual, acoustic and parent rated measures. All studies include an author closely linked to the method and institute.

Breathing, phonation and speech rate were the focus in four studies with in total 53 participants between 5 and 18 years old [47-50]. All four studies reported improved voice quality and speech intelligibility at the end of intervention for most participants. Follow-up after seven weeks showed a decreased effect after treatment termination [47].

EPG was used in three studies with a total of nine participants between 7 and 15 years [51-53]. One study included children with traumatic brain injury (TBI) [53]. All three studies report significant improvement according to perceptual assessment and/or analysis of EPG-data [51-53]. Morgan and coworkers (2007) found no or limited generalization effects [53]. No generalization effects were reported in the other two studies.

PROMPT was used in two studies with in total twelve participants between 3 and 11 years old [54,55]. Reported results were improved jaw-control and lip rounding in all participants and in 5/6 also improved speech intelligibility and performance of motor speech movement patterns.

Oral myofunctional treatment or Non-speech Oral Motor Treatment (NSOM), was used to improve lip-, tongue- and jaw-control in 16 children 7 to 10 years old [56]. The intervention consisted of movement and target training for the tongue, lips, and jaw muscles to establish adequate posture and function. Results showed significant improvement of lip-, tongue- and jaw-control and articulation of single words. Improved tongue control correlated with improved articulation.

One study described results after Phonetic placement therapy (PPT) combined with surface electro-myography feedback (sEMG) for a 13-year-old child [57]. The intervention consisted of three stages: baseline, PPT treatment and finally sEMG treatment separated by a 2-week no treatment period. Results revealed significant improvement in single word intelligibility following PPT with improvements maintained following sEMG treatment. The sEMG facilitated biofeedback in relaxation treatment. Perceptually, there was no change to any parameters of articulation following either treatment.

Scientific support for oral sensory-motor intervention methods for children and youth with cerebral palsy or traumatic brain injury and dysarthria based on assessed intervention studies are:

- *Medium high scientific support* for LSVT LOUD.
- *Limited scientific support* for EPG, Breathing, phonation and speech rate, Oral myofunctional treatment and PROMPT due to low number of participants, conflicting results in studies using the same method and other factors affecting study quality.

**Table 3:** Studies reporting treatment outcomes for children and adolescents with cerebral palsy (CP) or traumatic brain injury (TBI) and speech disorders. Authors, intervention methods, study design, age and number of included participants, type of disorder, additional disabilities if reported and GRADE is presented.

| Reference                             | Intervention                      | Participants | Diagnosis/Disorder |                           | GRADE |
|---------------------------------------|-----------------------------------|--------------|--------------------|---------------------------|-------|
|                                       |                                   | Sample size  | Age, y             |                           |       |
| Boliek, Fox, 2017 <sup>46</sup>       | LSVT LOUD                         | 7            | 6-10               | CP                        | 3     |
| Fox, Boliek 2012 <sup>43</sup>        | LSVT LOUD                         | 5            | 5-7                | CP                        | 3     |
| Boliek, Fox, 2014 <sup>44</sup>       | LSVT LOUD                         | 2            | 10                 | CP                        | 2     |
| Levy et al., 2012 <sup>45</sup>       | LSVT LOUD                         | 3            | 3:03, 8:10, 9:7    | CP, CAS                   | 1     |
| Pennington et al., 2006 <sup>47</sup> | Breathing, phonation, speech rate | 6            | 10-18              | CP                        | 2     |
| Pennington et al 2010 <sup>48</sup>   | Breathing, phonation, speech rate | 16           | 12-18              | CP                        | 2     |
| Pennington et al., 2013 <sup>50</sup> | Breathing, phonation, speech rate | 15           | 5-11               | CP                        | 2     |
| Miller et al., 2013 <sup>49</sup>     | Breathing, phonation, speech rate | 16           | 12-18              | CP                        | 2     |
| Nordberg et al., 2011 <sup>51</sup>   | EPG                               | 5            | 7:4-13:9           | CP                        | 2     |
| Gibbon, Wood, 2003 <sup>52</sup>      | EPG                               | 1            | 8                  | CP, phonological disorder | 1     |
| Morgan et al., 2007 <sup>53</sup>     | EPG                               | 3            | 14:10-15:01        | TBI                       | 1     |
| Ward et al., 2013 <sup>54</sup>       | PROMPT                            | 6            | 3-11               | CP                        | 3     |
| Ward et al., 2014 <sup>55</sup>       | PROMPT                            | 6            | 3:6-11:9           | CP                        | 3     |
| Ray, 2001 <sup>56</sup>               | Oral myofunctional treatment      | 16           | 7-10               | CP                        | 2     |
| Marchant et al., 2008 <sup>57</sup>   | PPT;sEMG                          | 1            | 13                 | CP                        | 1     |

**Abbreviations:** GRADE: Grading of Recommendations Assessment Development and Evaluation; CP: Cerebral Palsy; CAS: Childhood Apraxia of Speech; EPG: Electro Palato Graphy; TBI: Traumatic Brain Injury; PROMPT: Prompts for Restructuring Oral Muscular Phonetic Targets; PPT: Phonetic Placement Therapy; sEMG: surface Electro-MyoGraphy

**Table 4:** Motor learning principles in recommended methods based on Maas et al. (2008).

|                                | ReST<br>n=4          | DTTC<br>n=7                      | PROMPT<br>n=6            | Breathing &<br>phonation<br>n=4  | Palatal plate<br>n=5               | EPG<br>n=6                | LSVT<br>n=4            | MSTP<br>n=2            | NDP3<br>n=1          |
|--------------------------------|----------------------|----------------------------------|--------------------------|----------------------------------|------------------------------------|---------------------------|------------------------|------------------------|----------------------|
| Total no of participants       | N=24                 | N=19                             | N=27                     | N=53                             | N=294                              | N=18                      | N=17                   | N=42                   | N=13                 |
| No of sessions/w               | 4                    | 1-daily                          | 1-2 + home-work          | 3-5                              | Daily (≥2 t/d)                     | 2-5 + homework            | 4 + home-work          | 1-2                    | 4                    |
| Total no of sessions           | 12                   | 9-50                             | 14-40                    | 15-40                            | 16-≥2000                           | 14-40                     | 16                     | 10-20                  | 12                   |
| Length of session in minutes   | 60                   | 30-60                            | 45                       | 20-40                            | < 30                               | 15-60 + 10-15 min at home | 60 + 5-15 min at home  | 45                     | 60                   |
| Distributed vs mass practice   | Mostly distributed   | Mostly distributed               | Mass and distributed     | Mass practice in five blocks     | Passive or active use, distributed | Mostly distributed        | Mass and distributed   | Distributed            | Mostly distributed   |
| Blocked vs randomized practice | Mostly randomized    | Mostly randomized                | Blocked and randomized   | Blocked                          | Mostly blocked                     | Blocked                   | Blocked and randomized | Mostly randomized      | Mostly randomized    |
| Who performed the therapy?     | SLT or SLT stud      | SLP, teacher, caregiver          | Caregiver, SLP           | SLT or SLT stud                  | Caregiver, SLP                     | Caregiver, SLP            | Caregiver, SLP         | SLP                    | SLT stud             |
| Where was therapy performed?   | Clinic, school       | Clinic, school, home             | Home, clinic             | Clinic                           | Home, clinic                       | Home, clinic              | Home, clinic           | Clinic                 | Clinic               |
| Feedback                       | Following a schedule | Performance and result; fade out | Randomized and immediate | Performance and result; fade out | Sensory/tactile                    | Visual                    | Immediate; fade out    | Performance and result | Following a schedule |
| Generalization effects         | Yes                  | Yes                              | Yes                      | Varied -unsure                   | Unsure                             | Varied                    | Varied                 | Yes                    | Yes                  |
| Follow-up*                     | 4 m                  | 3 m                              | 1 y                      | 7 w                              | 4 y                                | 6 m                       | 3 m (12 w)             | 2 w                    | 4 m                  |

\*Longest reported follow-up for the method.

For other described intervention methods, the scientific support was found insufficient.

### Methods for children and youth with dysarthria due to neuromuscular disease

Only one study was identified describing results after intervention for children and youth with dysarthria due to neuromuscular disease. The study described results after treatment with an oral screen in eight children and adolescents between 7 and 19 years with dysarthria due to myotonic dystrophy [58]. Results showed improvement in 7/8 participants regarding lip muscle strength, 4/8 showed significant improvement. However, improved lip-muscle strength did not automatically lead to improved speech production.

No recommendations are made due to the very limited number of studies and low number of participants.

### Therapy structure and motor learning principles related to recommended methods

Identified motor learning principles for recommended methods are listed in table 4. Shared aspects for these methods were individualized and intense treatment with up to four sessions in the clinic/week (ReST, DTTC) or daily treatment provided by a parent or teacher under the supervision of a speech and language pathologist/therapist (SLP/SLT) (DTTC, PROMPT, LSVT LOUD, palatal plate, EPG), including many repetitions of the desired movements or targets (ReST, DTTC, PROMPT, LSVT LOUD, palatal plate, EPG).

A few studies specifically investigated effects related to intervention design. Maas and Farinella (2012) studied effects of blocked versus random practice on treatment performance and transfer in children with CAS [23]. Results related to practice design varied in the study group. Skelton and Hagopian (2014), reported substantial effects of randomized variable practice in children with CAS [25]. Edeal and Gildersleeve-Neumann (2011) investigated effects related to production frequency in speech therapy in two children to determine whether more practice leads to increased performance within a session, as well as improved motor learning and generalization to untrained words [19]. The higher production treatment required 100+ productions in 15 min, while the moderate treatment required 30–40 productions. Both children showed improvement on all targets. The targets with the higher production frequency were acquired faster as evidenced by better in-session performance and greater generalization to untrained probes. All three studies investigated effects related to DTTC.

Namasivayam and coworkers (2015) investigated effects related to treatment intensity in children with CAS using MSTP [30]. Results indicated that treatment twice/w during ten weeks yielded significantly better outcomes for articulation and functional communication compared to once/w.

The total number of sessions varied between studies from 12 to over 2000. The highest number is estimated based on recommendations in a four-year longitudinal study of palatal plate therapy for children with DS [39]. Studies including children with syndromes, cerebral palsy or acquired brain injury often

report a higher number of treatment sessions for improvement [28-29,59].

Generalizing treatment to non-treated items is an important sign of treatment effect [8,16,60]. Generalization to non-treated items were reported for *ReST*, *DTTC*, *PROMPT*, *MSTP* and *NDP3*. For *LSVT LOUD* varying generalization effects were found for both vocal loudness and speech accuracy. Limited generalizations were also reported following intervention with *palatal plates* and *EPG*.

## DISCUSSION AND CONCLUSION

This review evaluated the scientific evidence for oral sensory-motor interventions for children and adolescents with dysarthria, CAS or SSD. For children with CAS, one method, *ReST*, showed *high scientific support* based on four studies, including one RCT and a total of 24 treated children and adolescents, all indicating positive effects following the intervention. This effect also continued after treatment had ended which could be interpreted as an indication that a reorganization of motor commands and coordination had occurred. In a recently published Cochrane review the outcome from the RCT was assessed [61]. They concluded that both investigated interventions, *ReST* and *NPD3*, can be recommended based on results from this rigorous RCT study [16]. In the present review *ReST* was rated higher than *NPD3* based on a larger number of published studies and stronger evidence for long term effects.

In children with dysarthria and DS *limited to medium high scientific support* was found for interventions using palatal plates based on two RCT studies [38-39] and one group study [40]. This intervention and follow-up requires a multi professional team including dentist and SLP. Both speech and oral motor targets improved in all three studies. In the four-year follow-up [39] the difference between the treated and untreated group was less apparent possibly indicating a habituation, and thus less activation when using the palatal plates. Effects related to treatment duration for this group needs to be investigated further.

For children with cerebral palsy *LSVT LOUD* was given *medium high scientific support*. The scientific support for *EPG*, *Breathing*, *phonation* and *speech rate*, *Oral myofunctional treatment* and *PROMPT* were rated as *limited scientific support*. All studies included a low or fairly low number of participants. After the termination date for inclusion, one study reporting positive results following *LSVT LOUD* in children with CP have been published [62]. Previously reported positive results were corroborated by diffusion tensor imaging (DTI) indicating increased connectivity and white matter changes, strengthening the result in the present review. The most recent Cochrane review on intervention for children with CP did not result in any recommendations since no RCT studies were identified [6]. For children with dysarthria due to neuromuscular disease the scientific support is insufficient due to the limited number of identified studies.

Number of treated individuals for each method is low for most methods and varies between 1 and 294 ( $\bar{x}=30.67$ ). Several studies lacked a control group or showed other methodological limitations. Most studies report positive effects on one or

several aspects of speech production or functions underlying speech production, following intervention. In a meta-analysis, observational studies showed a tendency to report larger treatment effects than RCT studies [63].

The minimum intensity showing effects for children with CAS were two sessions a week [17,30]. Methods also including home training are typically directed to children with more severe disorders or disorders including cognitive impairment [28-29,38-39]. Home training increases the dose and may improve maintenance and generalization of acquired skills [21,26]. How skills are maintained long-term in patients with cognitive impairment remains unknown and needs to be further investigated. In the Carlstedt studies [38-39] children with DS were treated with palatal plates during a four-year period. After four years, speech and communication was similar to the untreated group [39].

Only a few of the included studies described how feedback was provided. Usually this is in the form of systematic feed-back schemes starting with feedback on performance, then feedback on results followed by delayed feedback and gradual fade-out to increase patient control (Table 4).

Dose or number of trials/session has been mentioned as an important factor particularly for children with CAS where an increased number of trials may be needed to induce change [59]. However, a high number of trials/session is reported for several of the included methods. Both ReST, DTTC, LSVT and PROMPT include large number of trials/session. In the literature, generally increased amount of practice is reported to lead to a better result [64] and maybe also greater retention. Depending on the amount of practice and the active participation needed to obtain improvement in several of the studied methods it may also be important to communicate the needed commitment to patients, caregivers and others involved in the intervention. The effort should be weighed against other simultaneous interventions and the child's own acceptance, understanding and motivation. Motivation is especially important in intensive training schemes [21] or schemes extending over a long period of time.

Before any intervention can be recommended, several questions need to be considered and discussed with patient/caregiver. The clinician should deliberate on different methods, is one preferred and if so why? Does the patient, caregivers or others involved have necessary resources and motivation to carry out the intervention? To answer these and similar questions the clinician need to have knowledge about the evidence base for different intervention methods for a specific dysfunction and patient group. The final decision is made in collaboration between clinician, patient and/or a caregiver.

Speech disorders affect quality of life and later occupational status in children with speech and language disorder. A 20-year follow-up of young adults with and without a history of speech and language impairment showed poorer outcomes in multiple domains including occupational status compared to peers without early communication impairment [65]. Thus, improving knowledge on effective and efficacious intervention methods is important.

## CONCLUSION

ReST was the only method with high scientific support for children with CAS. DTTC, PROMPT, NDP3 and MSTP had medium high scientific support in the treatment of children and adolescents with CAS and for PROMPT also in interventions for children and adolescents with SSD. LSVT LOUD had medium high evidence for children with dysarthria and cerebral palsy. The most apparent motor learning principle in recommended methods was high frequency intervention with 2 sessions/week or more. Children with syndromes had longer reported treatment periods. There is a need for high quality intervention studies, especially for children and adolescents with motor speech disorders and DS, CP, TBI and neuromuscular disease.

## CONFLICT OF INTEREST

All the authors declare no conflict of interest. This work was supported by "Föreningen för Sveriges Habiliteringschefer", Sweden, <http://habiliteringsverige.se/>

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