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Speech Intervention Effects for Childhood Apraxia of Speech:
Quality Appraisal of Systematic Reviews

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Review Article Submission to Perspectives
Abstract

Purpose: A number of studies have examined the effects of speech interventions on outcomes in childhood apraxia of speech (CAS). The findings have been summarized in the form of systematic reviews (SRs) and meta-analyses (MAs), which are used to support evidence-based clinical practice decisions. Yet without acceptable rigor, SRs/MAs may be biased in their recommendations. We appraised the quality of existing SRs for CAS treatment using a tool developed within epidemiology, the AMSTAR-2 (A MeaSurement Tool to Assess systematic Reviews; Shea et al., 2017).

Method: A search of five databases to identify published SRs that coalesced treatment research for CAS revealed six systematic reviews that met inclusion criteria. Two examiners coded each article with the AMSTAR-2 to rate the methodologic rigor of the SRs and extracted summary data.

Results: One rigorous systematic review included only one randomized controlled trial (Morgan et al., 2018). A second moderately rigorous review (Murray et al., 2014) examined multiple single participant research designs. The weight of high-quality evidence supported the positive effects of motor programming treatments for children with CAS.

Conclusions: Findings of six systematic reviews, two of which were conducted with relative rigor, suggest that motor programming treatments have the best evidence-base for treatment decisions pertaining to CAS. Clinicians are referred to online resources to implement these treatments according to published protocols.

Keywords: childhood apraxia of speech (CAS), systematic review (SR), evidence-based practice (EBP), research quality
Introduction

Childhood apraxia of speech (CAS) is an impairment evident when children struggle to develop articulate speech. Children with CAS have difficulty forming speech sounds during word production, especially as words increase in syllable length, producing distortions of vowels and sound substitutions, voicing errors, and schwa insertions, and using a slow rate with equal stress across syllables (Shriberg et al., 2017). A consensus definition of this challenging disorder identified CAS as an articulation impairment of neurologic origin, specifically impacting the planning and programming of movements for speech (American Speech-Language-Hearing Association, 2007). Current best diagnosis of CAS is made by expert opinion, relying on diagnostic features of inconsistent sound errors, difficulty with coarticulatory transitions between sounds and syllables, and inappropriate prosody (ASHA, 2007; Shriberg et al., 2017). Children with CAS demonstrate corresponding difficulties in activities of daily living (Tükel, Björelius, Henningsson, McAllister, & Eliasson, 2015) and systematically poorer language and academic outcomes than their peers (Gillon & Moriarty, 2007; Lewis, Freebairn, Hansen, Iyenger, & Taylor, 2004). The emotional and functional impact of CAS on the child and caregivers is often large (Miron, 2010).

Fortunately, several successful interventions for CAS have been developed and research support for them is growing. Among the approaches are interventions that incorporate motor-programming methods to improve articulatory skills, linguistic methods to enhance phonological skills, or alternative communication methods (ASHA, 2007). Examples of motor-programming approaches include Dynamic Temporal and Tactile Cuing (DTTC; Strand, Stoeckel, & Bass, 2006) and Rapid Syllable Transition Treatment (ReST; McCabe, Murray, Thomas, & Evans, 2017). In DTTC, clinicians provide integral stimulation cues to look, listen, and repeat
articulatory targets, typically to establish a core vocabulary of single words (Maas & Farinella, 2012; Strand et al., 2006). In contrast, ReST is an approach that emphasizes intensive practice with rapid syllable productions when reading multisyllable nonwords (Ballard, Robin, McCabe, & McDonald, 2010; McCabe et al., 2017). An example of a linguistic approach to treatment for CAS is phonological awareness training where children learn to segment, manipulate, and produce speech sound sequences (Moriarty & Gillon, 2006). Finally, those with severe CAS may need the support of augmentative and alternative communication methods to circumvent the considerable motor speech limitations (Cumley & Swanson, 1999).

Clinicians searching for the most effective way to treat their clients with CAS need an efficient way to identify the strongest evidence-based practices. Lacking time and access to complete and relevant literature resources, one useful approach to support evidence-based practice is for the clinician to identify systematic reviews or meta-analyses aligned with practice needs. Systematic reviews represent a rigorous process of identifying and synthesizing the results of the breadth of research studies on a topic, including an analysis of the methodologic quality of the research evidence (Grant & Booth, 2009). Some systematic reviews that use consistent outcome measures across studies incorporate a quantitative synthesis of the research evidence in the form of a meta-analysis. Most systematic reviews include all studies on a topic to encompass both the quantity and quality of evidence (Gough, 2007). While some advocate systematic review practices that combine only results from studies that meet quality guidelines (What Works Clearinghouse, 2017), such an approach maintains quality at the expense of ignoring the breadth of available evidence. Regardless of the approach, systematic reviews serve as valued documents to support evidence-based clinical practice decision-making.
To promote rigor in the conduct and reporting of systematic reviews, an international group developed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement (Liberati et al., 2009; Moher et al., 2009). The PRISMA checklist includes 27 elements intended to ensure a thorough search for all literature on a specified topic and use of sound methods to assure appropriate synthesis of the results and discussion of findings. Yet, the PRISMA authors emphasize that the checklist is not to be used to appraise the quality of systematic reviews and meta-analyses. Despite these guidelines, versions of which were developed as early as 1999 (Moher et al., 1999), systematic reviews vary in quality and can be biased in their conclusions if a rigorous systematic review method and synthesis is lacking (Schlosser, Wendt, & Sigafoos, 2007).

To that effect, a number of instruments have been developed to examine the rigor and lack of bias in the conduct of systematic reviews and meta-analyses. Within speech-language pathology, instruments aligned with these recommendations include the Evidence in Augmentative and Alternative Communication (EVIDAAC) Systematic Review Scale (Schlosser et al., 2008), and the Critical Appraisal of Systematic Review or Meta-Analysis (CASM; Dollaghan, 2007). Neither instrument has yet been thoroughly validated for use in appraising the quality of systematic reviews and meta-analyses. The most common instrument advocated for use in epidemiology circles to appraise the quality of SRs/MAs is A MeaSurement Tool to Assess systematic Reviews, recently revised as AMSTAR-2 (Shea et al., 2017). AMSTAR-2 was developed to appraise systematic reviews throughout healthcare-related fields that can encompasses both randomized and non-randomized treatment studies, allowing for an assessment of risk of bias in the research evidence.
As the incidence of CAS has increased over the last decades (ASHA, 2007), the number of systematic reviews addressing treatment research conducted for CAS has also grown. The purpose of this project was to evaluate the quality of SRs summarizing CAS treatment outcomes using the AMSTAR-2, a recognized quality appraisal tool, and to guide clinicians to the best research evidence available for intervention with CAS.

Method

Systematic reviews relevant to CAS published up to 2018 were located with three complementary processes. First, literature databases PsychInfo, PubMed, and Cochrane Database of Systematic Reviews were searched using the terms “childhood apraxia of speech” or “developmental apraxia of speech,” and “intervention” or “treatment” or “management”. Databases curated by national organizations for speech language pathology, ASHA Compendium (USA) and SpeechBite (Australia), were subsequently searched under the childhood apraxia of speech category. Finally, a hand-search of article references was also conducted. Articles were included if they were systematic reviews or meta-analyses that examined behavioral speech-language treatments for children with apraxia of speech. They were excluded if they did not contain CAS studies, did not incorporate behavioral treatments, included only adults, or were published in a language other than English. Two raters examined the resulting titles and abstracts of potential articles for inclusion and reached consensus agreement on the selection of reviews that met inclusion criteria. Seven review articles meeting inclusion criteria initially were identified for review and data extraction. Upon further reading, one article was removed (Allen et al., 2017) because treatment results for participants with CAS could not be distinguished independent of studies pertaining to other developmental disabilities, leaving six systematic reviews for quality appraisal.
Quality Appraisal and Data Extraction

Quality appraisal was conducted with the sixteen questions of the AMSTAR-2 checklist (Shea et al., 2017) to address the methodological rigor of the six identified systematic reviews. The AMSTAR-2 questions examine literature search and selection, data extraction and summary, summary of bias, and meta-analysis methods, if appropriate. Eleven questions are scored with one point if the feature is present. The remaining five questions allow for partial credit of one-half point. To receive a full point for a quality feature, relevant information must be explicitly stated in the published document. Review quality ratings were reported as percentages of the 16 AMSTAR-2 points available. Confidence in the review results was then determined in accordance with the critical domains recommended in Shea et al. (2017), referring to seven of the criteria that are especially important to reduce bias in a systematic review.

Two raters reviewed the AMSTAR-2 checklist and then together coded one article (Morgan & Vogel, 2009) to train on the use of the checklist. The raters then independently coded the other five papers on appraisal criteria. Following independent rating, disagreements between raters were discussed until consensus was reached. The inter-rater agreement was calculated for the remaining five articles and achieved a 90% agreement level. The percent of the 16 criteria met was calculated for each study. A modest 50% benchmark was identified to note the reviews that were conducted with relative rigor.

Finally, the characteristics of each systematic review article were recorded, including: number of studies in the review, study designs included, method of study quality rating, number of participants, diagnosis and age range of participants, general treatment approaches reviewed, treatment schedule, and summary of treatment outcomes.
Results

Six systematic reviews of interventions for CAS were located (Kaipa & Peterson, 2016; Koehlinger, 2015; Morgan, Murray, & Liegeois, 2018; Morgan & Vogel, 2009; Murray, McCabe, & Ballard, 2014; Roper, 2007). A descriptive summary of the results of the six reviews are available in Table 1 and quality appraisal results using the AMSTAR-2 are noted in Table 2. None of the reviews conducted a meta-analysis.

Review Characteristics

Table 1 shows that the number of studies summarized in the systematic reviews varied widely among the six articles, from zero (Morgan & Vogel, 2009) to 42 studies (Murray et al., 2014). The variation in number of studies was due in part to the study designs accepted in the inclusion criteria for the systematic reviews. Two reviews sought only randomized controlled trials (RCT) or quasi-RCTs (Morgan et al., 2018; Morgan & Vogel, 2009), one focused on single subject design (SSD) studies (Koehlinger, 2015), one included both SSD and case studies (Murray et al., 2014), one included SSD and quasi-experimental group studies (Kaipa & Peterson, 2016), and one included primary studies of all research designs (Roper, 2007). Morgan and Vogel located no studies that met the inclusion criterion of only randomized controlled trials, whereas Murray et al. reviewed 42 studies because they examined 23 SSDs and 19 case studies.

The total number of participants with CAS included in the systematic reviews ranged from zero (Morgan & Vogel, 2009) to 83 (Murray et al., 2014). All studies included children in the age range of three to eight years (Kaipa & Peterson, 2016; Koehlinger, 2015; Morgan et al., 2018; Morgan & Vogel, 2009; Murray et al., 2014; Roper, 2007). One study included children as young as 2 years, 8 months (Roper, 2007), one extended the range of participants to age 14 (Murray et al., 2014), and two more extended to the age of 16 (Kaipa & Peterson, 2016; Morgan
et al., 2018). Four of the systematic reviews limited study participants to CAS diagnosis only (Kaipa & Peterson, 2016; Koehlinger, 2015; Morgan et al., 2018; Morgan & Vogel, 2009), while the remaining two recorded data for children with CAS and co-morbid diagnoses (Murray et al., 2014; Roper, 2007).

**Treatments and Outcomes**

Table 1 displays that two reviews summarized multiple types of treatments, including linguistic, motor-programming, and alternative and augmentative communication approaches (Koehlinger, 2015; Murray et al., 2014). Two reviews included only motor-programming treatments: DTTC, ReST, and the Nuffield Dyspraxia Programme – 3 (Kaipa & Peterson, 2016; Morgan et al., 2018). One review was limited to Melodic Intonation Therapy effects for CAS (MIT; Roper, 2007). The schedule for reviewed treatments varied by study, but most identified hour-long sessions delivered one to three times weekly.

Table 1

Intervention outcomes reported among the systematic reviews varied from number of errors to intelligibility of verbal responses to accuracy of phoneme targets during the production of words, phrases, and connected speech. Five of the six reviews reported improvement in speech production following treatments for CAS. In a review of single subject designs, calculation of improvement rate difference showed large effect sizes for two motor programming treatments (DTTC, ReST) and medium effects for a phonemic awareness treatment (Murray, McCabe, & Ballard, 2014). Better outcomes were reported for high intensity motor programming treatment schedules that applied a high dose (number of responses) and higher dose frequency (days per week) during training (Kaipa & Peterson, 2016). No outcomes were reported in Morgan and Vogel (2009) because no randomized controlled trials were identified for review.
Their follow-up review in 2018 (Morgan et al., 2018) identified one RCT that showed improvements for two motor-programming treatments for CAS.

Quality of Review

Ratings on the AMSTAR-2 (Shea et al., 2017) criteria are shown in Table 2. Seven criteria essential for high confidence in the review findings, six of which pertain to systematic reviews and one to meta-analyses, are shaded in the table. The percentage of AMSTAR-2 criteria addressed for each review is listed in Table 1. The most recent review by Morgan and colleagues (2018) using the Cochrane systematic review protocol addressed all criteria essential for an unbiased systematic review and addressed 68.8% of the 16 AMSTAR-2 criteria. All other reviews lacked at least one of the six essential criteria. The review by Murray and colleagues (2014) demonstrated a moderate level of rigor in addressing 43.8% of the AMSTAR-2 criteria, yet omitted an assessment of risk of bias that is deemed essential in a systematic review. The remaining four reviews demonstrated a low level of rigor, ranging 6.3-18.8% of AMSTAR-2 criteria met.

Table 2

Table 2 details the scores for each systematic review for each AMSTAR-2 criterion. The most frequently met quality criterion (4 of 6 reviews) was reporting of funding sources and potential conflicts of interest. Most (3 of 6 reviews) engaged in a comprehensive literature search and study selection, justified the exclusion of studies, and considered risk of bias in the interpretation of findings from the original research studies, all of which are considered critical domains of the AMSTAR-2. Three reviews also completed study selection in duplicate and provided a satisfactory explanation of the heterogeneity of their findings. In contrast, no reviews addressed meta-analysis components of the AMSTAR-2.
Discussion

A number of research groups have conducted systematic reviews of the treatment research literature for CAS. For clinicians to appreciate the conclusions of those systematic reviews, it is important to appraise the quality of methods conducted in those reviews. Familiarity with some of the criteria included in a tool such as AMSTAR-2 can be helpful to guide the interpretation and confidence in the conclusions of a systematic review.

Quality of Evidence

Among the six reviews that were identified in this critical appraisal project, the quality of methods varied considerably. The included reviews demonstrated clear strengths: they conducted comprehensive literature searches with study selection duplicated across investigators, included basic information on excluded studies with justification, considered risk of bias within the primary studies, explained the heterogeneity of results, and revealed funding for the review. Only one of six included reviews was completed with high rigor on the AMSTAR-2, the review completed by Morgan et al. (2018) within the Cochrane Collaboration. Yet that review summarized only one treatment study that met the inclusion criterion for randomized controlled trials. One other moderately-well-conducted systematic review by Murray and colleagues (2014) summarized findings from 42 studies and calculated effect sizes for the 23 single subject designs among the 42 studies.

None of the systematic reviews completed a meta-analysis of findings, which would allow a higher level of confidence due to the statistical aggregate of findings. Yet the heterogeneity of outcome measures implemented across original studies undermines the ability to calculate meta-analysis metrics. The combination of results from different types of primary
studies can increase both the statistical complexity of meta-analysis and the risk of bias within the review (Shea et al., 2017).

**Treatment Methods**

General agreement among the systematic reviews indicates that reduced articulation errors and increased intelligibility follows CAS treatment. Although there is some indication of positive results from the linguistic treatment method of integrated phonological awareness (Murray et al., 2014), evidence for other types of linguistic and augmentative and alternative communication methods is limited. The best systematic reviews appraised in this project, Murray and colleagues (2014) and Morgan and colleagues (2018), used different search criteria but came to complementary conclusions supporting motor programming treatments (e.g., Integral Stimulation, DTTC, ReST, the Nuffield Dyspraxia Programme). Integral Stimulation and DTTC demonstrated a moderate treatment effect and ReST demonstrated a large effect (Murray et al., 2014). Although the systematic review methods are somewhat lacking, Kaipa and Peterson (2016) report that motor programming treatments administered with higher dose and higher dose frequency were found to be more effective. Overall, motor programming treatments that focus training at the syllable or word level appeared to be among the best documented in the literature and most effective approaches to improve verbal production in CAS.

**Implications for Clinical Practice and Future Directions**

Although the evidence from large RCTs is lacking (Morgan et al., 2018), the preponderance of evidence including less rigorous study designs summarized in recent systematic reviews (e.g., Murray et al., 2014), suggests that motor programming treatments lead to the best speech production outcomes in CAS. Among these approaches are Rapid Syllable Transition Treatment (ReST; McCabe et al., 2017), Dynamic Temporal and Tactile Cueing
(DTTC, Strand et al., 2006), and Nuffield Dyspraxia Program (Murray, McCabe, & Ballard, 2015; Williams & Stephens, 2010). Descriptions of these treatment approaches can be found in various online resources (e.g., apraxia-kids.org; asha.org; sydney.edu.au/health-sciences/rest/; ndp3.org/). Additional professional training can be beneficial to effectively administer these treatment protocols and are available through these and other websites (e.g., childapraxiatreatment.org; utdallas.edu/calliercenter/events/CAS/; sydney.edu.au/health-sciences/rest/training-package/). Some supporting treatment materials and training are provided by the researcher/developers free of charge, while others require purchase.

The current project also highlights the need for future efforts to document the effects of CAS treatments in rigorous controlled research designs, thereby adding to the evidence base of future Cochrane systematic reviews. Among the six reviews examined in this project, none conducted a meta-analysis, which is not surprising given the array of outcomes tested across original studies. Identification of a standard set of outcome measures to be used across clinical trials would support future efforts to coalesce the research using meta-analysis. Finally, future systematic reviews would benefit from careful consideration of documents developed in epidemiology circles, such as the PRISMA statement (Moher et al., 2009) and the AMSTAR-2 (Shea et al., 2017), to assure thorough, unbiased methods and recommendations. Meantime, clinicians will use current evidence from original studies and systematic reviews to guide the selection of the best treatments possible for their clients with CAS, focusing primarily on motor programming treatment approaches.
References


doi:10.3109/17549507.2015.1010578


<table>
<thead>
<tr>
<th>Authors, Date</th>
<th>AMSTAR-2 score</th>
<th>Number of Studies Reviewed</th>
<th>Method of study Quality Ratings</th>
<th>Diagnosis of subjects</th>
<th>Design of Treatment studies included</th>
<th>Age Range (years)</th>
<th># Subjects</th>
<th>Type of Treatments Reviewed</th>
<th>Schedule of Treatments (in general)</th>
<th>Outcomes/Conclusions</th>
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</thead>
<tbody>
<tr>
<td>Roper, 2007</td>
<td>18.8</td>
<td>3</td>
<td>None</td>
<td>CAS; autism</td>
<td>All</td>
<td>2.75-8</td>
<td>6</td>
<td>Melodic Intonation Therapy</td>
<td>1-3x/wk for 2-33 months</td>
<td>Decreased articulation errors: 3/3 studies; Increased intelligibility for target phrases: 3/3 studies; Increased naming: 2/3 studies; Increased repetition: 1/3 studies; Increased spontaneous words &amp; MLU utterances: 1/3 studies; Evidence is “meager”; studies affected by threats to internal validity and multiple treatment interference</td>
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<tr>
<td>Morgan &amp; Vogel, 2009</td>
<td>31.3</td>
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<td>NA</td>
<td>CAS</td>
<td>RCTs, Quasi-RCTs</td>
<td>3-16</td>
<td>0</td>
<td>NR</td>
<td>NR</td>
<td>Preponderant evidence: 7 pts Integral Stimulation/Dynamic Temporal &amp; Tactile Cueing: Improvement rate difference (IRD)=.60 moderate effect; 15 pts</td>
</tr>
<tr>
<td>Murray, McCabe, &amp; Ballard, 2014</td>
<td>43.8</td>
<td>42</td>
<td>Phase of research</td>
<td>CAS or CAS+</td>
<td>23 SSD, 19 Case Studies</td>
<td>3-14</td>
<td>83</td>
<td>SSDs: 11 motor; 6 linguistic with some motor; 4 linguistic with</td>
<td>Median 3x/wk 2-5 days/wk</td>
<td>Preponderant evidence: 7 pts Integral Stimulation/Dynamic Temporal &amp; Tactile Cueing: Improvement rate difference (IRD)=.60 moderate effect; 15 pts</td>
</tr>
<tr>
<td>Authors, Date</td>
<td>AMSTAR-2 score</td>
<td>Number of Studies Reviewed</td>
<td>Method of study Quality Ratings</td>
<td>Diagnosis of subjects</td>
<td>Design of Treatment studies included</td>
<td>Age Range (years)</td>
<td># Subjects</td>
<td>Type of Treatments Reviewed</td>
<td>Schedule of Treatments (in general)</td>
<td>Outcomes/Conclusions</td>
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<td>Koehler, 2015</td>
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<td>Integrated Phonemic Awareness intervention: IRD=.51 moderate effects; n=3 pts Rapid Syllable Transition: IRD=.78 large effect Descriptive: Greatest evidence available for motor treatments delivered with high production frequency and lower feedback with visual and verbal feedback</td>
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<td>Kaipa &amp; Peterson, 2016</td>
<td>18.8</td>
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<td>CAS</td>
<td>1 group</td>
<td>NS</td>
<td>43 CAS</td>
<td>Several included REST and motor speech treatment</td>
<td>Higher intensity vs Lower intensity</td>
<td>High dose and higher dose frequency led to better outcomes</td>
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<td>Morgan, Murray, &amp; Leigeois, 2018</td>
<td>68.8</td>
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<td>CAS</td>
<td>RCTs &amp; Quasi-RCTs</td>
<td>3-16</td>
<td>26</td>
<td>Nuffield dyspraxia program-3 vs. Rapid syllable transition</td>
<td>4 1-hr sessions/wk for 3 wks</td>
<td>Both treatments increased outcomes at 1 month post-treatment for treated and untreated words and connected speech</td>
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Table 2: Scores on each AMSTAR-2 (Shea et al., 2017) quality criterion; shaded items are typically essential to reduce bias.

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<td>8-Included studies described in detail</td>
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<td>14-Satisfactory explanation of heterogeneity</td>
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<td>15-MA publication bias analysis/impact on results</td>
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Note: PICO = population, intervention, comparison, outcome; MA = meta-analysis; NA = not applicable. Shaded lines indicate critical domain components, as recommended by Shea et al. (2017). Items in **bold font** were frequently identified in at least half of the included SRs. Items in *italics* apply only to meta-analysis and are not applicable to any of the included SRs.
Continuing Education Questions (5)

*** indicates correct response

1. A diagnostic feature that suggests the presence of CAS includes:
   a. Word retrieval difficulties
   b. Poor selection of plural morphemes
   c. Difficulty with coarticulatory transitions between sounds and syllables ***
   d. Normal prosody

2. A systematic review that combines outcome measures across studies in a quantitative synthesis of the research evidence is a:
   a. Meta-analysis ***
   b. Meta-synthesis
   c. Quantifiable review
   d. Aggregate summary

3. This systematic review of CAS treatment identified one randomized controlled trial that met their inclusion criteria:
   a. Morgan and Vogel (2009)
   b. Murray, McCabe, and Ballard (2014)
   d. Morgan, Murray, and Leigeois (2018)***
4. The AMSTAR-2 appraisal found that most of the systematic reviews of CAS treatment included the following component:
   a. Conflict of interest/Funding source ***
   b. Assessed risk of bias for included studies
   c. Completed data extraction in duplicate
   d. Used appropriate statistical combination of results

5. Strongest research evidence exists to support this treatment approach for CAS:
   a. Language production treatment
   b. Augmentative and alternative communication approaches
   c. Motor programming treatment ***
   d. Phonemic awareness training

Learning Outcomes (3)

After reading this manuscript, clinicians will be able to:

1. State three treatment approaches often used for children with apraxia of speech.
2. Indicate seven characteristics of rigorous systematic reviews.