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Review Article

Treatment for Acquired Apraxia of Speech: A Systematic Review of Intervention Research Between 2004 and 2012

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Objectives: The aim was for the appointed committee of the Academy of Neurological Communication Disorders and Sciences to conduct a systematic review of published intervention studies of acquired apraxia of speech (AOS), updating the previous committee's review article from 2006.
Method: A systematic search of 11 databases identified 215 articles, with 26 meeting inclusion criteria of (a) stating intention to measure effects of treatment on AOS and (b) data representing treatment effects for at least 1 individual stated to have AOS.
Results: All studies involved within-participant experimental designs, with sample sizes of 1 to 44 (median = 1). Confidence in diagnosis was rated high to reasonable in 18 of 26 studies. Most studies (24/26) reported on articulatory–kinematic approaches; 2 applied rhythm/rate control methods. Six studies had sufficient experimental control for Class III rating according to the *Clinical Practice Guidelines Process Manual* (American Academy of Neurology, 2011), with 15 others satisfying all criteria for Class III except use of independent or objective outcome measurement.
Conclusions: The most important global clinical conclusion from this review is that the weight of evidence supports a strong effect for both articulatory–kinematic and rate/rhythm approaches to AOS treatment. The quantity of work, experimental rigor, and reporting of diagnostic criteria continue to improve and strengthen confidence in the corpus of research.

Acquired apraxia of speech (AOS) is a motor speech disorder that is typically caused by stroke. It can range from minimal or mild to complete loss of the ability to speak. In some cases, AOS resolves quickly in the acute phase following stroke or neurological injury, but for many people it is a chronic condition that significantly affects communication in everyday situations.

AOS is a disruption in spatial and temporal planning and/or programming of movements for speech production. There is wide consensus that AOS is characterized by slowed speech rate with distorted phonemes, distorted phoneme substitutions, and a tendency to segregate speech into individual syllables and equalize stress across adjacent syllables (Duffy, 2013; McNeil, Robin, & Schmidt, 2009). Although AOS can involve all speech subsystems, it is predominantly a disorder of articulation and prosody. It is important to note that although there is agreement on these features, there is ongoing debate regarding their relative contribution to a diagnosis and the importance of other features, such as errors perceived as undistorted phoneme substitutions or consistency of errors over repeated productions (e.g., Staiger, Finger-Berg, Aichert, & Ziegler, 2012).

Research examining behavioral interventions for AOS has focused primarily on identifying the presence of a treatment effect and developing replicable treatment protocols. This has generated a corpus of literature reflecting single-case experimental designs, case series, and uncontrolled case studies. According to the *Clinical Practice Guidelines Process Manual* developed by the American Academy of Neurology (AAN, 2011), the majority of these studies represent Class III to IV. Research categorized as Class III includes controlled studies (e.g., within-participant experimental designs) where confounding factors contributing to differences

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between groups are described, and masked, objective, or independent assessment of the primary outcome measures are used. Class IV studies use uncontrolled research designs that may not provide clear evidence that participants meet specific diagnostic inclusion criteria, do not sufficiently define interventions, or use biased outcomes measures.

Class III studies are an essential first step in investigating treatment efficacy, and inevitably precede treatment efficacy research meeting criteria for Class I to II (e.g., randomized controlled trials or group-level case-control studies). Given the current status of research in treatment efficacy for AOS, it is not surprising that a Cochrane Review of randomized controlled trials investigating speech outcomes for AOS poststroke concluded that there were no suitable studies for review (West, Hesketh, Vail, & Bowen, 2005).

The first systematic review on this topic, sponsored by the Academy for Neurological Communication Disorders and Sciences (ANCDS) was completed in 2006 (Wambaugh, Duffy, McNeil, Robin, & Rogers, 2006a, 2006b). It included all research identified from a systematic search and reviewed a broader range of research comprising all levels of evidence demonstrating some scientific rigor. Studies were evaluated for features such as internal and external validity, reliability of dependent and independent measures, and sufficient methodological detail to allow replication by independent researchers.

In that review, Wambaugh and colleagues (2006a, 2006b) analyzed 59 publications from 1951 to 2003 and concluded that individuals with AOS can benefit from behavioral intervention. Of the 59 studies, only two represented group-level studies, with just one of them being an experimental group study. The majority of the studies comprised within-participant experimental designs, case series, and uncontrolled case studies (i.e., Class III or IV). Consistent with this type of research, the majority of studies had small sample sizes. The following observations about participant characteristics and interventions were reported. A total of 146 participants were studied. Close to 80% were men, and close to 20% were women, 93% presented with an etiology of stroke, and 94% were diagnosed with moderate or severe AOS. Only 17.5% of studies provided sufficient description of participant characteristics to support confidence in a diagnosis of AOS. Young adults (i.e., under 65 years) were overrepresented. Studies were grouped into categories based on the focus of the intervention protocol, with some studies incorporating components of more than one category: articulatory–kinematic (n = 30 studies), prosody (specifically, rate and/or rhythm; n = 7), alternative or augmentative communication (AAC; n = 8), intersystemic reorganization (i.e., multimodal training; n = 8), and other (n = 5). With AOS being defined as a phonetic-motoric disorder, just over half of the published studies aimed to improve articulatory–kinematic skills.

More recently, Strom (2008) completed quantitative meta-analyses of eight group and 11 single-subject design studies reporting outcomes for articulatory–kinematic treatments using measures of correct words or sounds. Findings indicated that, for both single-case and group study designs, AOS treatment is efficacious. Despite this conclusion, Strom cautioned that overall improvements in speech output may be small and the analyses were likely influenced by publication bias, that is, "the tendency for authors to submit, and of journals to accept, manuscripts for publication based on the direction or strength of the study findings" (Hopewell, Loudon, Clarke, Oxman, & Dickersin, 2009, p. 2).

Here, we present a systematic review using the same format applied by Wambaugh et al. (2006a, 2006b) to allow direct comparison of findings from the current and previous reviews. The previous review included plans to provide an update to the original systematic review when approximately 30 new treatment investigations had been conducted or 5 years had elapsed (Wambaugh et al., 2006b).

Purpose

The purpose of the current review was to examine intervention studies intending to treat acquired AOS that have been published since the completion of the original ANCDS systematic review in 2006. Through this process we identify advances in the field that will guide evidence-based practice. The review adheres to the PRISMA guidelines for conducting systematic reviews (Liberati et al., 2009). For each study reviewed, the following study components were evaluated to establish the strength of the studies and, therefore, their potential to contribute to the evidence base for guiding clinical practice:

- The scientific adequacy of each study in terms of sample size, evidence of internal and external validity, and calculations of reliability for dependent and independent variables
- The description of study participants and resulting degree of confidence in the diagnosis of AOS
- The description of the treatment applied, including whether sufficient methodological detail was provided for independent study replication, the type of intervention approach, the dependent measures selected, treatment dose, use of specific principles of motor learning (PML), and potential bias in the dependent measures
- The measurement of the treatment effect, including measurement of treatment and response or stimulus generalization effects, maintenance of treatment effects, stability of untreated behaviors for demonstration of experimental control, and any measurement of a treatment's social/ecological validity

Method

The protocol for evaluating the studies, developed for the 2006 review, was revised for this review and was approved by the ANCDS Steering Committee on Evidence-Based Practice Guidelines. All steps outlined in the PRISMA statement relevant to reporting systematic reviews were applied (Liberati et al., 2009). The review committee was appointed by the ANCDS chair of Professional Practice

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and Practice Guidelines and comprised current members of the ANCDS who have published on diagnosis and/or treatment of acquired AOS. Several members of the initial AOS Guidelines Writing Committee continued to serve on this reconstituted committee to provide continuity. The committee included the seven authors, all of whom have extensive research and clinical experience with AOS diagnosis and treatment. The committee completed all stages of the review.

Evidence Identification

A systematic literature search was conducted to identify studies reporting treatment outcomes for behavioral interventions for AOS. The following 11 databases were searched: CINAHL via EBSCOhost; Cochrane Database of Systematic Reviews; Embase; Expanded Academic ASAP; Google Scholar; MEDLINE; PubMed; ProQuest; ProQuest Dissertation and Thesis (PQDT); ScienceDirect; Scopus; and Web of Science.

Targeted search terms using Boolean operators were applied as follows: *Apraxia AND (Speech OR Communication) AND (Treatment OR Treatment outcome OR Therapy OR Intervention OR Rehabilitation)* in key word/topic/subject heading search options. Variations were used per database operating systems (e.g., wildcard variation using *). Additional search terms suggested by a database were accepted if relevant (e.g., *therapeutics*). Where possible, searches were limited to adult populations or included in the Boolean search with the addition of the search terms *(aged OR adult)*. Similarly, in order to effectively search for the most relevant articles, modifiers regarding etiology (e.g., *brain injury OR stroke*) were added as additional search terms where limiters were not available.

The search was limited to articles published in English. The search dates were restricted to January 2003 to December 2012, overlapping with the final year of the previous review to ensure detection of any articles that might have been overlooked. To minimize overlooking relevant articles missed in searches, reference lists of identified articles were checked. No additional articles were identified.

Screening

Following removal of duplicate articles, the search produced 215 articles. All articles were analyzed for relevance to the clinical question. Two reviewers independently screened all titles and abstracts and discarded 171 articles because they did not meet inclusion criteria. Specifically, the articles initially excluded from the review reported on etiologies that varied from sudden-onset acquired AOS, including progressive AOS, childhood AOS, acquired communication disorders other than AOS (e.g., dysarthria or aphasia), or other apraxias (e.g., limb apraxia, ideational apraxia). Articles not reporting treatment outcomes (i.e., outcomes from intervention applied over multiple sessions) and treatment reviews with no new experimental data were also excluded at this point.

Eligibility Criteria

Three reviewers scanned the remaining 44 articles to ensure they met inclusion criteria for formal review. Studies were required to report measurement of the effects of treatment on behaviors attributed to AOS at any level of impairment of function. Specifically, articles were required to include (a) a statement on intention to treat AOS or intention to measure the effects of treatment on AOS, and (b) data representing treatment outcomes (positive or negative) for at least one individual stated to have AOS. An additional 17 articles were excluded for the following reasons: They were published conference abstracts with insufficient detail for fair analysis; they did not include a statement of intention to treat AOS or measure treatment effects pertaining to AOS; or they were doctoral dissertations superseded by published papers. Four disagreements between reviewers regarding inclusion were resolved by consensus. One study was retained but combined with another study by the same authors because it represented additional outcome measures from the one experiment; therefore, the two studies were treated as a single study (Youmans, Youmans, & Hancock, 2011a, 2011b). Excluded articles were not analyzed further. Finally, bibliographies of included articles were scanned for any additional studies; none were identified. Details of the number of studies identified and screening procedures used to generate the final 26 articles included in this review article are shown in Figure 1.

Rating the Evidence

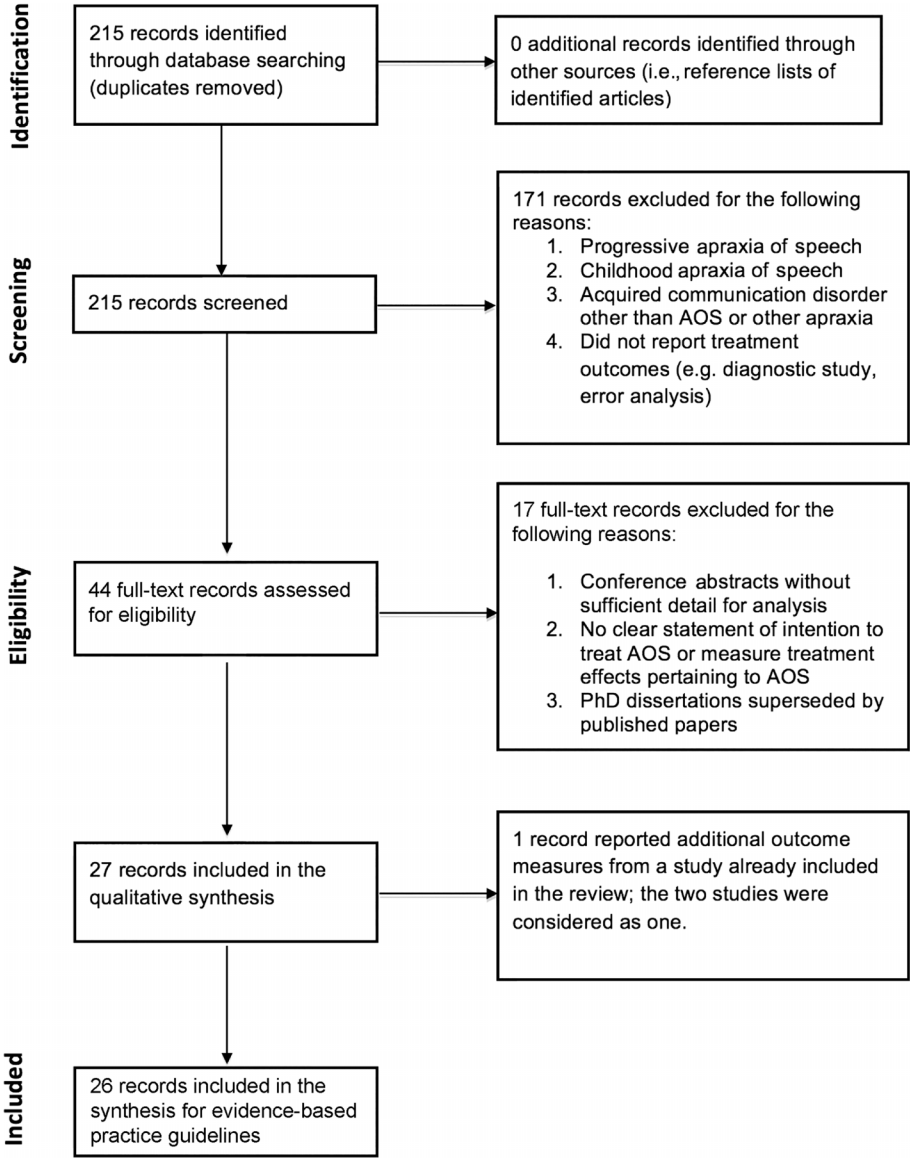
The 26 articles identified for full review were rated using the criteria developed for the 2006 review (Duffy & Yorkston, 2003; Wambaugh et al., 2006a; Yorkston et al., 2001). A Microsoft Excel spreadsheet was created for raters to enter their ratings and text comments for each article. The articles were distributed among the committee members, ensuring that raters were not assigned articles on which they were an author. Raters were not blinded to each article's title or authors. To ensure reliable ratings by the committee, (a) all articles were rated independently by two authors on the Single-Case Experimental Design (SCED) scale (Tate et al., 2008) or modified Physiotherapy Evidence Database scale (PEDro-P; Murray et al., 2013); (b) for all remaining variables, 14 articles (54%) were rated independently by two authors, and, because interrater reliability was high (i.e., 86%–96%), the remaining 12 articles were rated by a single rater. For double-rated articles, discrepancies in ratings were resolved by two-thirds consensus using a third author prior to inclusion in the final evidence table. Original percentage agreement scores are reported in the relevant sections of Results.

Scientific Adequacy

Ratings of scientific adequacy considered sample size, evidence of internal and external validity, calculations of reliability for dependent and independent variables, and presentation of sufficient methodological detail for independent

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Figure 1. Flow of information through the systematic review.



study replication. First, an AAN (2011) classification was assigned to each study to indicate level of evidence, with Class I being randomized controlled clinical trials; Class II being a retrospective cohort study or a case-control study otherwise meeting the criteria for Class I or a randomized controlled trial lacking one to two criteria for a Class I rating; Class III being controlled studies including within-participant designs that use masked, objective, or independent outcome assessors; and Class IV being uncontrolled studies or studies with no clear diagnostic criteria for participant inclusion or inadequately defined outcome measures. Because 15 of the 26 studies reviewed here satisfied all criteria for Class III but did not define masked, objective, or independent outcome assessment, we marked these studies as Class III-b; the writing committee unanimously agreed that these studies demonstrated sufficient rigor to justify assignment to Class III rather than Class IV. For the remaining 11 studies, six attained a Class III rating and five, a Class IV rating (see Results).

Where possible, studies were also rated on a scale designed to evaluate methodological bias in the research design. For studies that used single-case experimental design, the SCED scale (Tate et al., 2008) was used to assign a score out of 1 to 10 indicating methodological quality. The scale involves binary ratings of 11 design features that, when present, reduce bias: description of clinical history and target behaviors; robust design; multiple baselines; sampling of primary dependent variables during the treatment phase; indication of variability in the data set; calculation of interrater reliability; independent assessment of dependent variables;

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statistical analysis to support interpretation; replication across participants, clinicians, or communicative contexts; and demonstration of response and/or stimulus generalization. The clinical history item is not used in computing the SCED score.

For any studies judged to be group experimental trials, the PEDro-P scale (e.g., Murray et al., 2013) was used to assign a score of 1 to 10 indicating methodological quality. Similar to the SCED scale, the PEDro-P scale rates articles on the presence of items associated with reduced methodological bias for group studies. For the PEDro-P scale, these items include avoidance of pretreatment biases with random and concealed allocation of participants to treatment conditions as well as statistical verification of comparability of participants at baseline; blinding of participants, therapists, and assessors to the treatment conditions and the study hypotheses to control performance biases during the study, biases associated with data analysis including dropout rates, the use of intention-to-treat analysis, and the rigor of statistical reporting (Maher, Sherrington, Herbert, Moseley, & Elkins, 2003).

All 26 rated investigations received two independent ratings on either the SCED scale or PEDro-P scale. Authors C.L. and K.B. completed primary ratings for all articles. The second raters were four students in a graduate seminar in single-subject design under the direction of author J.W. Two students independently rated each report, and any disagreements were resolved by author J.W. prior to calculating reliability with the primary rater.

Description of Study Participants

Basic demographic information was extracted from each article including age, severity of AOS, presence of concomitant communication disorders, etiology, method of diagnosis, and participant inclusion/exclusion criteria. Critically, the level of confidence in the authors' diagnosis of AOS was rated using a modified three-level version of the five-level rating scale developed by Wambaugh et al. (2006a). This scale uses the definition and diagnostic feature list for AOS developed by McNeil, Robin, and Schmidt (1997). To accommodate recent debate over some features (e.g., consistency of error type and location over repeated productions of words), we collapsed the top three ratings (1, 2, 3) into a single A rating (diagnosis of AOS based on all or most of the features of AOS listed, with no or few listed features also present in aphasia), representing reasonably high confidence in AOS diagnosis. We retained the two lower levels as B (incomplete or inadequate description AOS features) and C (no description of features). Each article was scanned for evidence that participants demonstrated the widely agreed-upon features of AOS, namely, slow speech rate due to protracted segments and intersegment durations, phoneme distortions or distorted phoneme substitutions, and dysprosody (e.g., equalization of stress across syllables and syllable segregation). The modified scale did not include the feature of consistency in error type and location, accommodating recent studies showing that this behavior is heavily dependent upon the task characteristics and stimuli used (Haley, Jacks, & Cunningham, 2013; Mauszycki, Wambaugh, & Cameron, 2012; Scholl & Ballard, 2012; Staiger et al., 2012). Although additional features may have been listed in each study, such as articulatory groping or speech initiation difficulties, many have convincingly argued that these features do not discriminate AOS from other concomitant disorders (McNeil et al., 2009).

Description of Treatment

For each intervention, several attributes were rated as present or absent and a brief description was generated (see Table 1). These included presence of sufficient methodological detail for faithful replication or meaningful cross-study comparisons; intervention type recorded as articulatory–kinematic, rate and/or rhythm, or other; primary dependent measure(s); frequency and duration of intervention sessions; and duration of the treatment program. Application of any PML was also recorded to reflect the shift in motor learning studies toward the PML framework since the last review (Bislick, Weir, Spencer, Kendall, & Yorkston, 2012; Maas et al., 2008; Schmidt & Lee, 2011).

To identify any risks of bias, each study was examined for blinding of those collecting and/or analyzing the outcome data and for loss of participants to follow-up. For group studies, reviewers also noted evidence of random and concealed allocation to groups. It is acknowledged that participants and treatment providers cannot be blinded to the treatment condition.

Measurement of Treatment Effects

To evaluate the quality and scope of measurement of treatment effects, information was extracted on the dependent measures used to test (a) change in treated behaviors, (b) change in untreated related behaviors (i.e., generalization of treatment effects to different stimuli or responses), (c) change in untreated unrelated behaviors (i.e., control behaviors), and (d) maintenance of treatment effects at 2 or more weeks posttreatment (McReynolds & Kearns, 1983). Any measures of social/ecological validity were documented. For group studies, reviewers were asked to note evidence of intention-to-treat analysis.

Expert Reviewers

A draft of this review article was distributed for comment to members of the writing committee, ANCDS office bearers, and corresponding authors of all studies included in the review article as well as other scientists who have published in the area of AOS. A total of 26 reviewers provided comment. The draft report was revised after careful consideration and discussion of all comments among all authors of this review article.

Results

For the 26 AOS intervention studies included in the systematic review, a comprehensive table of ratings was generated. This table was used to then generate a series of summary tables, presented in this section, to aid interpretation

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Table 1. Summary of participant details, treatment techniques and targets, design standard, and reported treatment effects for the 26 studies included in the review.

First author (year)	Sample size	Diagnostic rating	AOS severity	Treatment techniques and targets	AAN class[a]	Reported treatment effects
Articulatory–kinematic approaches						
Aitken Dunham (2010)	2	B	Severe +aphasia ?dysarthria	Speech and language treatment: 8-step task continuum and naming with a hierarchy of cues from nil to imitation Targets: single words Music therapy: Singing familiar songs; slow and gentle breathing of CVC syllables; producing novel phrases using familiar melodies, loudness variations, and pauses during singing with and without clapping; and oral-motor exercises during singing of familiar songs Targets: Songs and novel phrases	IV	Improvement on treated speech and naming skills and standardized assessments following both treatment approaches; greatest treatment effects following the combined speech-language and music treatment.
Austermann Hula (2008)	Exp 1: 4 Exp 2: 2	A	Mild to severe +aphasia 1 dysarthria	Prepractice: phonetic placement strategies using orthographic stimuli, articulatory placement diagrams, verbal descriptions of articulatory features, and modeling Practice: Exp 1: alternated low- (60%) and high- (100%) frequency knowledge of results feedback, provided immediately Exp 2: alternated immediate and 5-sec-delayed knowledge of results feedback, provided with high frequency Targets: 1- to 3-syllable nonwords with singletons and/or 2- to 3-consonant clusters, depending on AOS severity [b]PML comparison	III-b	Exp 1: 2 of 4 participants showed the predicted benefit of low-frequency feedback for maintenance. Exp 2: 1 of 2 participants showed the predicted benefit of delayed feedback for maintenance. Other factors such as stimulus complexity, task difficulty, and AOS severity may have influenced the response to the feedback conditions.
Ballard (2007)	2	A	Moderate +aphasia –dysarthria	Prepractice: phonetic placement strategies using orthographic stimuli, articulatory placement diagrams, verbal descriptions of articulatory features, and modeling plus spectographic feedback on timing of voicing Practice: random practice of items, fading knowledge of results feedback provided with a 3- to 4-s delay Targets: CVC real words with initial voiced and voiceless phonemes PML structure	III-b	Both participants showed some degree of improvement with treatment, maintenance, and generalization to related words, but findings need to be replicated in a larger sample.
Cowell (2010)	6	B	Mild to severe +aphasia ?dysarthria	Self-administered computer programs: (a) speech treatment—multimodality sensory stimulation including auditory, visual, orthographic, visual object, somatosensory, and "imagined" production, followed by actual word production; (b) sham visuospatial treatment—short-term memory pattern recognition tasks and timed jigsaw completion Targets: Single words	III-b	Improvements in word accuracy and duration; improvements for speech treatment greater than the sham visuospatial exercises regardless of treatment order. Rising baselines in both treatment conditions complicated interpretation.

(*table continues*)

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Table 1 *(Continued).*

First author (year)	Sample size	Diagnostic rating	AOS severity	Treatment techniques and targets	AAN class[a]	Reported treatment effects
Davis (2009)	1	A	Not reported +anomia –dysarthria	Implicit phoneme manipulation tasks (i.e., no overt speech during treatment) with computer-based tasks for rhyming, alliteration, and deletion of sounds in words Targets: single words	III	Improved overt speech production with increased accuracy of target sounds in treated words (i.e., reduction in sound distortions, phonological errors, and improved prosody); generalization of effects to improved production of target sounds to untreated words supported by both visual inspection and effect-size data. Differential effects of the three treatment techniques cannot be determined. Design would be improved with more than two baseline tests and continued testing to obtain maintenance data for the final sound-target. Subsets of data, rather than all data, appear to be selectively presented.
Friedman (2010)	1	A	Moderate–severe ?aphasia –dysarthria	Motor learning guided approach[c] Step 1: Unison production, immediate repetition, reading aloud, answering questions in random order; Step 2: Modeling and reading aloud with repetitions Step 3: Home practice of reading aloud and repetitions, modeling via audiotape as needed, no augmented feedback PML: Summary and fading knowledge of results feedback provided with a delay, knowledge of performance provided as needed, stimuli presented in randomized blocks. Targets: 3- to 5-word functional phrases PML structure	III-b	Production accuracy and response time improved with treatment; however, some improvement may have been related to increasing familiarity with the probe task and apparent nonblinding of raters. Interrater reliability for scoring of responses was low at 70%.
Katz (2010)	1	A	Not reported +aphasia ?dysarthria	Augmented visual feedback on tongue position (EMA) during imitation and reading aloud PML: Contrasted high-frequency visual feedback on all trials with low-frequency visual feedback on a random 50% of trials Targets: CVC real words PML comparison	III-b	Visual feedback resulted in improved production for some but not all treated targets. Contrary to prediction, high-frequency feedback resulted in stronger maintenance than low. Generalization to untreated words was evident. Interpretation complicated by high variability on the control behavior and small number of stimuli.
Kendall (2006)	1	B	Moderate +aphasia ?dysarthria	Phonomotor rehabilitation: Lindamood Phoneme Sequencing program[d] modified to incorporate PML; imitation, articulatory placement diagrams, auditory discrimination tasks, articulatory contrast tasks, tactile/kinesthetic cues, visual feedback via	III	Production of isolated phonemes improved. Posttreatment, discourse production was judged to be less effortful but slower and less natural sounding. The participant showed a high level of accuracy with words of

(*table continues*)

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Table 1 *(Continued).*

First author (year)	Sample size	Diagnostic rating	AOS severity	Treatment techniques and targets	AAN class[a]	Reported treatment effects
				mirror, mental practice via identification of correct articulation of phonemes from pictures and auditory models PML: random presentation of varied stimuli, mental practice, high-frequency knowledge of performance feedback, intermittent immediate knowledge of results feedback Targets: isolated phonemes PML structure		increasing length prior to treatment, questioning the rationale for targeting isolated phonemes.
Lasker (2008)	1	B	Severe +aphasia ?dysarthria	Motor learning guided approach with imitation, immediate and delayed repetition, reading aloud, home practice using a speech-generating device to generate models for imitation PML: stimuli presented in randomized blocks, delayed and summary knowledge of results feedback at low frequency (~30%) Targets: functional CV words, progressing to 2-syllable then 3- to 7-syllable words and phrases PML structure	IV	A feasibility study suggesting that this approach may be effective for severe AOS. The study outcomes are difficult to interpret due to lack of multiple baselines for comparison with treatment data, loss of experimental control with untreated targets improving during treatment, concurrent participation in an aphasia group, and mainly anecdotal evidence for maintenance.
Lasker (2010)	1	B	Severe +aphasia ?dysarthria	Motor learning guided approach with imitation, immediate and delayed repetition, reading aloud, sessions conducted face-to-face and via Skype, home practice using a speech-generating device to generate models for imitation PML: stimuli presented in randomized blocks, delayed and summary knowledge of results feedback at low frequency (~30%) Targets: 4- to 12-syllable phrases PML structure	IV	Speech production showed improvement. Behavioral changes during Skype-delivered treatment lagged slightly behind face-to-face treatment.
Marangolo (2011)	3	A	Severe +aphasia ?dysarthria	Behavioral treatment: Imitation with cuing hierarchy; modeling with segregation of syllables, vowel prolongation, and exaggerated articulatory gestures, modeling one syllable at a time Anodic tDCS stimulation: 1mA current for 20 min, electrodes placed over inferior frontal gyrus (BA44/45) and contralateral supraorbital region during behavioral training Sham stimulation: Same as anodic stimulation but stimulator turned off after 30 s by independent operator Targets: nonword CV, real-word CVCV and CVCCV with different place and manner represented	IV	All participants showed greater response accuracy with anodic vs. sham stimulation in at least one testing condition (e.g., word repetition, word reading). Results transferred across one or more related tasks for all participants. Treatment effects were maintained for the tDCS stimulation condition only. Replication is required in a larger sample, preferably with between-groups methodology.

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Table 1 *(Continued).*

First author (year)	Sample size	Diagnostic rating	AOS severity	Treatment techniques and targets	AAN class[a]	Reported treatment effects
McNeil (2010)	2	A	Mild–moderate +aphasia ?dysarthria	Repeated practice of words with online visual kinematic feedback via EMA plus verbal feedback PML: Contrasted high-frequency visual feedback on all trials with low-frequency visual feedback on a random 50% of trials Targets: Initial and final consonants in 1-syllable words PML comparison	III-b	Kinematic plus verbal feedback resulted in improved perceptual accuracy for most exemplars of the treated phonemes, with positive generalization to untreated related words for both participants. One participant maintained treatment effects. Effects of high vs. low feedback difficult to interpret.
Rose (2006)	1	A	Moderate +aphasia –dysarthria	Verbal treatment: Identifying phoneme errors, contrasting correct and incorrect productions, matching production with articulatory placement diagrams, imitation, orthographic cues, feedback via a response-contingent hierarchy Gesture treatment: As above, using gestural cues (i.e., cued articulation) in place of orthographic cues[e] Verbal plus gestural: As above, with both orthographic and gestural cues PML: multiple sounds of high complexity (i.e., clusters) targeted simultaneously, random stimulus presentation Targets: Single words with consonant singletons and clusters PML structure	III-b	All three treatment approaches led to significant improvement in production of treated words for one individual with chronic AOS. Treatment effects generalized to improved production of untreated stimuli as well as treated and untreated words in novel elicitation tasks. Given the simultaneous administration of three treatments within-subject, there is reasonable risk of contamination across treatments and so increased risk of Type II error.
Savage (2012)	1	B	Severe –aphasia +dysarthria	Phonetic placement therapy (pictured articulatory placement cues, tactile placement cues, auditory models) provided during conversations using AAC device when the participant's response contained one of the target sounds; high-intensity practice with each target elicited 40–50 times in 30 min. Targets: Single words	III	Single-word speech intelligibility improved from 0% to 24%.
Schneider (2005)	3	C	Moderate-severe +aphasia ?dysarthria	Modified 8-step continuum[f] with imitation, unison production, syllable by syllable production, and tactile and verbal articulatory instructions. PML: Contrasted training low- vs. high-complexity stimuli Targets: 4-syllable nonwords increasing in complexity from one consonant one vowel (e.g., *bobobobo*), to one consonant different vowels to different consonants one vowel to different consonants different vowels (*bonatigu*) PML comparison	III-b	Treatment resulted in improved production of trained nonwords for all participants. Consistent with prediction, generalization occurred to less complex nonwords but for only one participant. Generalization from less to more complex nonwords for one participant was unexpected.

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Table 1 *(Continued).*

First author (year)	Sample size	Diagnostic rating	AOS severity	Treatment techniques and targets	AAN class[a]	Reported treatment effects
van der Merwe (2007)	1	A	Not reported[g] –aphasia –dysarthria	Speech Motor Learning Program: Not described but see van der Merwe (2011) in next row of Table PML: blocked and random practice, variable practice, low-frequency feedback (type not specified), low- and high-complexity stimuli Targets: words and nonwords targeting easy and difficult phonemes and clusters in 1- to 3-syllable structures PML structure	III-b	With treatment, the total number of errors decreased, although this improvement began during baseline. Also, the number of attempted self-corrections decreased and number of successful self-corrections increased. However, the percentage of attempted self-corrections on incorrect productions remained stable, suggesting no improvement in processes of feed-forward motor control.
van der Merwe (2011)	1	A	Moderate –aphasia –dysarthria	Speech Motor Learning Program: progresses from imitated blocked practice of nonwords to self-initiated production of nonword series and real words, uses hierarchy from less to more complex stimuli, integral stimulation PML: random practice schedule, variable practice, self-monitoring, rate increases, delayed repetition, low- and high-complexity stimuli. knowledge of results feedback fading over the program Targets: words and nonwords targeting easy and difficult phonemes and clusters in 1- to 3-syllable structures PML structure	III	Treatment led to improvements in speech production; however, unexpected improvements often occurred in as-yet untreated behaviors, particularly for real words. This unexpected change represents a loss of experimental control, although pretreatment baselines showed no systematic improvement, and for some stages there was greater change once treatment began. The intervention appears complex, and it is not clear what aspects of the treatment are necessary or effective.
Waldron (2011)	1	C	Unclear +aphasia ?dysarthria	Three treatment techniques compared (a) auditory discrimination, (b) speech production and repetition with integral stimulation and articulatory placement cues, and (c) self-monitoring Targets: single words	IV	Authors argue that the production and monitoring treatments improved spoken naming, but auditory discrimination treatment did not. There were no improvements on reading and repetition. Some design features make interpretation of results difficult (e.g., unstable baselines, no withdrawal phase or probing of untreated behaviors; confounds of order and carryover effects, no reported reliability on dependent and independent measures).
Wambaugh (2004)	2	A	Mild to severe +aphasia –dysarthria	Sound production treatment: modeling, repetition, minimal pair contrast, integral stimulation, articulatory placement cues, verbal feedback via a response-contingent hierarchy Targets: 2-word phrases for both participants, /r/ blends in monosyllabic words for one participant.	III-b	Treatment resulted in increased accuracy of production of treated sounds/ phonemes in trained and untrained words under probe conditions. Effects generalized strongly to untrained exemplars of trained phonemes, replicating previous studies. Generalization to sentence-level contexts was weak, prompting authors to recommend direct treatment of these more complex contexts.

(*table continues*)

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Table 1 *(Continued).*

First author (year)	Sample size	Diagnostic rating	AOS severity	Treatment techniques and targets	AAN class[a]	Reported treatment effects
Wambaugh, Nessler (2004)	1	A	Moderate–severe +aphasia +dysarthria	Sound production treatment: modeling, repetition, minimal pair contrast, integral stimulation, visual cues, graphemic cues, verbal feedback via a response-contingent hierarchy Targets: specific phonemes in monosyllabic words	III-b	Positive acquisition and maintenance effects; positive generalization of trained phonemes to word-final position but weak generalization to production of treated items in a sentence completion task. An additional, controlled, treatment phase was applied to the sentence completion context, with positive effect.
Wambaugh (2012) (also included rate/rhythm approach)	10	A	Not stated +aphasia –dysarthria	Repeated practice: Repeating words 5 times each after a model, with summative feedback on accuracy Repeated practice with rate/rhythm control PML: high-intensity, summative feedback Targets: words or sound within words PML structure	III-b	Repeated practice resulted in positive treatment effects; addition of rate/rhythm control did not provide systematic benefit over the repeated practice alone; small additional gains noted for most but not all of rate/rhythm applications
Whiteside (2012)	44	A	Mild to severe group average in range of aphasia ?dysarthria	Self-administered computer programs: (a) speech treatment—multimodality sensory stimulation including auditory, visual, orthographic, visual object, somatosensory, and "imagined" production, followed by actual word production; (b) sham visuospatial treatment—short-term memory pattern recognition tasks and timed jigsaw completion Targets: single words	III	A significant reduction in struggle behavior and an increase in accuracy for trained and phonetically matched untrained words with speech treatment; only the group in speech treatment first showed continued reduction in struggle behavior during sham treatment, which authors attribute to a carryover effect. Speech treatment did not stimulate improvement in untreated frequency-matched words.
Youmans (2011a, 2011b)	3	A	Mild to severe +aphasia ?dysarthria	Script training: Step 1: Blocked practice of one phrase at a time, clinician modeling, unison production, reading aloud, independent production without cues Step 2: Random practice of independent productions with and without orthographic cues Home practice with audiotapes PML: random practice, summary feedback Targets: personalized dyadic scripts PML structure	III-b	Youmans (2011a; all 3 participants): Positive acquisition of trained scripts with maintenance of treatment effects. Participants reported using the scripts frequently, but this response generalization was not tested experimentally. Anecdotally, participants reported increased confidence, speaking ease, and speech naturalness. It cannot be determined whether application of PML influenced outcomes.

(*table continues*)

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Table 1 *(Continued).*

First author (year)	Sample size	Diagnostic rating	AOS severity	Treatment techniques and targets	AAN class[a]	Reported treatment effects
						Youmans (2011b; 1 of 3 participants): Social validity was examined with one of the three participants. More accurate and fluent script performance was judged by listeners to be more understandable and natural and produced with more ease. Overall, the speech sample from the treatment phase was perceived by naïve listener to be of better quality. However, it is unclear whether findings support the efficacy of script training because samples do not represent pre- and posttreatment points.
Rate and/or rhythm control approaches						
Brendel (2008)	10	A	Mild to severe 9 aphasia 3 dysarthria	Metrical pacing treatment: Production of a sentence in synchrony with a tone sequence, representing the rhythmic pattern of syllable onsets with attention to synchronizing rather than articulation; visual feedback showing the amplitude envelope of the production in relation to the tone sequence; feedback on rate, fluency, and pattern matching; additional cues (tapping, tactile cues, chorus-speaking) provided as needed Articulation treatment: Attention to articulatory accuracy using phonetic placement strategies, gestural facilitation, integral stimulation, minimal pairs, word derivation method Targets: words or phrases	**III**	Metrical pacing therapy produced improvements in suprasegmental aspects (sentence duration and proportion of dysfluencies) as well as in articulatory (segmental) accuracy, even though participants did not receive feedback or focus on the latter. Articulation treatment produced improvements in articulatory accuracy only. The improvements in articulation were statistically comparable for both treatments.
Mauszycki (2008)	1	A	Mild +anomia –dysarthria	Hand tapping and production one syllable at a time in synchrony with metronome, adjusted to individual speaking rate; clinician model, unison production; repetitions; attention to accuracy of rate/timing not articulation Targets: words in phrases	III-b	Improved sound production accuracy and total utterance duration in repetition tasks, even though sound production accuracy did not receive feedback or focus.

Note. AOS = apraxia of speech; AAN = American Academy of Neurology; Exp = experiment; PML = principles of motor learning; EMA = electromagnetic midsagittal articulography; tDCS = transcranial direct current stimulation; BA = Brodmann area; AAC = alternative or augmentative communication. Diagnostic Rating Scale: A = all or most of the features of AOS listed, with no or few listed features also present in aphasia, B = incomplete or inadequate description AOS features, C = no description of features; + or – aphasia or dysarthria indicates concomitant aphasia or dysarthria (unilateral upper motor neuron type in all cases except for Savage et al., 2012, who reported hypokinetic type) were present/absent, ? indicates no report of concomitance.

[a]AAN (2011): Level III includes controlled studies where participants serve as their own control; when groups are studied, baseline differences that might affect outcomes are described, and outcome assessment is masked, objective, or conducted by an independent assessor; Level III-b includes studies that satisfy all Level III criteria except masked, objective, or independent outcome assessment; Level IV includes studies that do not satisfy multiple Level III criteria. [b]For more on PML, see Schmidt and Lee (2011). [c]Hageman, Simon, Backer, and Burda (2002). [d]Lindamood and Lindamood (1993). [e]Passy (1990). [f]Rosenbek, Lemme, Ahern, Harris, and Wertz (1973). [g]Same individual reported in van der Merwe (2011).

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and comparison. Of the 26 studies analyzed, 24 applied a primarily articulatory–kinematic approach that aimed to improve speech sound accuracy. Two applied a primarily rhythm/rate approach without direct training of articulatory accuracy and monitored changes in utterance duration, fluency, and speech sound accuracy (see Table 1). In addition, one of the articulatory–kinematic studies included a rate/rhythm phase of treatment.

Scientific Adequacy

The studies varied in sample size from 1 to 44 participants with reported AOS (median = 1, lower quartile [LQ] = 1, upper quartile [UQ] = 3; i.e., 75% of studies had from one to three participants). Three studies had ≥10 participants (n = 10: Brendel & Ziegler, 2008; n = 10: Wambaugh, Nessler, Cameron, & Mauszycki, 2012; n = 44: Whiteside et al., 2012).

Most studies (21/26, 81%) were classified as AAN Class III (n = 6) or III-b (n = 15), indicating evidence of internal validity (i.e., some degree of experimental control was described, allowing reasonable confidence that the reported effects were due to the application of the treatment) (see Tables 1 and 2). The remaining five studies were classified as AAN Class IV, being uncontrolled studies and/or containing no clear evidence that participants met diagnostic criteria for AOS. Given that all 26 studies used within-participant designs, none qualified for an AAN Class I or Class II rating. All of these studies had participants serve as their own controls. Interrater agreement on assigning a III/III-b versus a IV rating was 100%.

Of the 26 studies, 21 were judged to use some form of single-case experimental design and were scored on the SCED scale. Average SCED score was 6.6 out of 10 (SD = 2.4, range = 4–9, median = 7). Two studies were judged as group-experimental studies and were rated on the PEDro-P scale. These received scores of 7 out of 10 (Whiteside et al., 2012) and 3 out of 10 (Brendel & Ziegler, 2008). The final three studies included a case report, a case series, and a small group study. These could not be rated using available validated methodological rating scales. All 23 rated investigations received two independent ratings on either the SCED scale or PEDro-P scale. For the 21 SCED-rated studies, the 11 items were rated for agreement (a total of 231 ratings) with point-to-point agreement at 96%. There were 10 disagreements within eight studies as follows: scoring of designs (3); scoring of baselines (2); scoring of replications (1); scoring of independent assessor (1); scoring of generalization (2); and scoring of statistics (1). Interrater reliability on the PEDro-P ratings was 86% (19/22 points rated). All disagreements were reviewed and a final rating was provided on the basis of consensus across the three raters K.B., J.W., and C.L.

The following outcomes were found when examining the studies from a methodological bias perspective. For the 21 single-case experimental design, studies all reported adequate clinical history and target behaviors, with over 90% reported adequate sampling and raw data records; 18 of 21 (85%) reported interrater reliability results; 17 of 21 (81%) reported using a robust research design and demonstrated generalization beyond the treatment targets; 15 of 21 (71%) reported stable multiple baselines pretreatment; and 12 of 21 (57%) reported replication across participants, therapists, or settings. Less than 50% (10/21, or 47%) of studies used statistical data analysis to interpret their findings. Only two of 21 (9%) reported the use of independent assessors to collect or analyze outcome data.

Of the two group-experimental studies that qualified for rating on the PEDro-P scale, both reported random allocation to treatment conditions; one reported concealed allocation and adequate baseline comparability. As is common with behavioral intervention, neither reported the use of blinded therapists or participants, but both used independent assessors of outcome data. Both reported statistical analyses to interpret their findings. Only one reported posttreatment outcomes for more than 85% of their participant sample, and neither reported using an intention-to-treat analysis.

Of the 24 studies reporting on articulatory–kinematic approaches, 20 used some form of single-case experimental design, two represented a case study or cases series, and two were group experimental studies incorporating within-participant comparisons (see Table 2). Thirteen tested the effects of a single treatment against a no-treatment baseline condition using multiple baseline designs across behaviors and/or participants. Four compared the effects of two or more specific principles of motor learning on the treatment's outcomes (i.e., feedback frequency, feedback delay, or stimulus complexity) using alternating treatments, ABACA design, or counterbalancing of conditions across

Table 2. Summary of study designs and internal validity by treatment type.

Type of treatment	Number of studies	Case series	Within-participant	Group-experimental	Evidence of internal validity[a]
Articulatory–kinematic	24	2	20	2	17 of 21
Rate/rhythm	2	0	1	1	2 of 2

[a]Credit given for design on the Single-Case Experimental Design (SCED) scale or between group/condition and baseline comparability on the Physiotherapy Evidence Database (PEDro-P) scale, that is, a design with sufficient experimental control to support the claim that treatment was responsible for the observed behavioral change. Of the three articulatory–kinematic studies that were not appropriate for rating with the SCED or PEDro-P scales, one of three showed evidence of internal validity.

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participants (Austermann Hula, Robin, Maas, Ballard, & Schmidt, 2008; Katz, McNeil, & Garst, 2010; McNeil et al., 2010; Schneider & Frens, 2005). Six directly compared two treatment approaches with alternating treatments or crossover design. Of these six treatment comparisons, one compared gestural and verbal treatments (Rose & Douglas, 2006), one compared articulation treatment with and without music therapy (Aitken Dunham, 2010), one compared articulation treatment with or without transcranial direct current stimulation over the left inferior frontal gyrus (Brodmann areas 44 and 45; Marangolo et al., 2011), one compared exercises focusing on either auditory or production tasks (Waldron, Whitworth, & Howard, 2011), and two compared articulation treatment versus a sham visuospatial condition delivered via computer (Cowell, Whiteside, Windsor, & Varley, 2010; Whiteside et al., 2012).

Of the three studies reporting on rhythm/rate approaches, one applied a group experimental design using crossover of treatment conditions in which all 10 participants received one phase of metrical pacing treatment and one phase with an articulatory–kinematic approach (Brendel & Ziegler, 2008). Another study applied a multiple-baselines design across behaviors with one participant and applied only rhythm/rate control intervention (Mauszycki & Wambaugh, 2008). A third study used a combined ABCA design (alternating treatments, multiple baselines across behaviors, and participants) in which rate/rhythm treatment was applied following repeated articulation practice treatment with eight participants (Wambaugh et al., 2012).

Study Participants

The 26 studies involved 107 participants with AOS aged between 28 and 87 years (M = 60 years), with an approximately 3:2 ratio of men (n = 63) to women (n = 44). In cases of multiple articles by a single research group, it is possible that some participants were involved in more than one study; this was only clear for the two studies by van der Merwe (2007, 2011; see Table 1).

The majority of cases were reported to have had a single left hemisphere middle cerebral artery stroke. Other etiologies included one case with multiple left hemisphere strokes (Lasker, Stierwalt, Hageman, & LaPointe, 2008), four with a basal ganglia lesion (two participants from Brendel & Ziegler, 2008; one each from Marangolo et al., 2011, and Savage, Stead, & Hoffman, 2012), one with right hemisphere middle cerebral artery stroke, one with encephalitic parkinsonism, one with traumatic brain injury due to gunshot (Friedman, Hancock, Schulz, & Bamdad, 2010), and one with traumatic brain injury[1] of unspecified cause (Ballard, Maas, & Robin, 2007). Participants ranged from 1 month to more than 20 years postonset, with the vast majority being in the chronic phase (i.e., >6 months postonset). Presence of concomitant aphasia was reported in 25 studies, with only three participants stated to have no aphasia. Presence of concomitant dysarthria was reported in 13 studies and, when present, was consistent with unilateral upper motor neuron dysarthria in all but one individual with hypokinetic dysarthria (Savage et al., 2012; see Table 1). Studies represented participants from Australia, Germany, Italy, South Africa, the United Kingdom, and the United States.

Regarding the information provided in each study to support the diagnosis of AOS, 18 of 26 (69%) scored 1 to 3 (i.e., an A-level rating), supporting high confidence in presence of AOS (see Table 1). That is, authors based the diagnosis of AOS on all or most of the characteristics associated with AOS and none or few characteristics that are considered nondiscriminative between AOS and aphasia or dysarthria. Six (23%) were rated as B, having incomplete descriptions that did not allow confidence in diagnosis. Two (8%) were rated as C because an AOS diagnosis was stated but no description of characteristics supporting the diagnosis was provided. Interrater reliability for rating diagnostic confidence level was 93% (13/14), with disagreement solved by two-thirds consensus using a third rater.

Description of Treatment

The intensity of treatment varied widely across studies (see Table 3). For studies in which number of sessions was clearly stated (20/26), the median total number of sessions was 28.5, ranging from two to 91 sessions (LQ = 12.8, UQ = 36.5). The median number of sessions per week was three, ranging from one to five sessions a week (LQ = 2, UQ = 3). The median total number of weeks for treatment delivery was 7.5, ranging from 1 to 78 weeks (LQ = 5, UQ = 16).

Fourteen of the 26 studies (54%) made some reference to incorporating PML. Four reported direct comparisons of one principle against another, whereas 10 reported using specific principles that are thought to promote long-term retention and generalization. This is coded in Table 2 as *PML comparison* or *PML structure* in the description of treatment techniques and targets for each study.

Measurement of Treatment Effects

Of the 21 studies rated as AAN Class III or III-b, 13 (62%) reported statistical analyses to support claims of positive treatment effects and/or positive maintenance or generalization of treatment effects (see Table 4). The most commonly used statistic was effect size, most often reported for individual participant data.

Of the 26 studies, 24 (92%) described primary dependent measures involving phonemic/phonetic accuracy. Additional measures included word or utterance duration, speech intelligibility, naming performance, signs of struggle or effortful speech, and instances of dysfluency. Apart from word and utterance duration, no study reported quantitative measures of prosodic features such as phrase-,

[1]This participant had sustained a focal head injury during a motor vehicle accident.

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Table 3. Summary of treatment amount and intensity by treatment type.

Type of treatment	Number of studies	Total number of sessions	Sessions per week	Total length of treatment
Articulatory–kinematic	24[a]	Median = 30 Range = 9–91	Median = 3 Range = 1–5	Median = 9.25 weeks Range = 2–78
Rate/rhythm	2	Median = 28.5 Range = 28–29	Median = 3 Range = 2–4	Median = 15.75 weeks Range = 7–25

[a]One study provided no information on treatment amount or intensity, four studies provided detail for only one variable, and one study provided detail for only two variables; these are included in the calculations.

word-, or syllable-level stress or duration and frequency of pauses or hesitations.

Measures of social/ecological validity were reported in just five studies (note that the two studies by Youmans et al., 2011a, 2011b, were combined for all other analyses). Measures included clinician ratings of the participant's communicative effectiveness when answering questions about daily activities (Brendel & Ziegler, 2008); independent clinicians' judgments pre- to posttreatment of speech rate, ease of speaking, naturalness, articulatory precision, and intelligibility during discourse and subjective evaluation by the participant via a questionnaire about ease of communication in daily activities (Kendall, Rodriguez, Rosenbek, Conway, & Rothi, 2006); anecdotal report about use of treated items in conversation (Lasker et al., 2008); self-ratings for confidence, ease of speaking, and naturalness during discourse (Youmans et al., 2011a); and judgment by 124 unfamiliar listeners of speech quality, intelligibility, naturalness, and ease of speaking during script productions sampled in the baseline and treatment phases of the study (Youmans et al., 2011b).

Discussion

In 2006, Wambaugh and colleagues (2006a) reviewed 59 studies reporting on efficacy of treatment for acquired AOS over the 33 years between 1970 and 2003. That report identified numerous weaknesses in the evidence base such as lack of experimental design, inadequate participant description, and minimal replication of treatment effects across participants and investigations. This review revealed an increase in the number of studies examining treatment efficacy in AOS. Here, we reviewed 26 additional studies that were published in the 10 years from 2004 to 2013 and demonstrate that considerable inroads have been made relative to these weaknesses.

Improvements in Scientific Adequacy

This latest generation of AOS treatment studies has added substantially to the existing body of literature supporting the use of articulatory–kinematic approaches in the treatment of AOS, with modest additional evidence provided for rate/rhythm approaches to improving speech production in AOS. The vast majority of reports in the 2006 guidelines aimed to detect and estimate the size of any treatment effect, categorized as Phase I investigations by Robey and Schultz (1998). This was not considered necessarily a weakness but rather a reflection of the immature state of the AOS treatment research. The quality of design has improved over these years, with almost all studies now using some degree of experimental control. In the current report, several investigations were designed to evaluate specific aspects of treatment (e.g., Austermann-Hula et al., 2008, Schneider & Frens, 2005; Wambaugh et al., 2012) or to compare treatment approaches (e.g., Brendel & Ziegler, 2008; Rose & Douglas, 2006; Waldron et al., 2011), classified as Phase II. However, studies still tended toward very small sample sizes, with more than half of the reports involving a single participant. This also hinders demonstration of replicable effects; the previous review article (Wambaugh et al., 2006a) noted that replication is a fundamental component of the scientific process but that lack of replication was a serious weakness in the AOS literature to 2003. Despite the large number of single-participant studies, replications across participants and investigations have occurred more frequently in this more recent group of studies.

Three studies had 10 or more participants, although these used within-participant designs comparing different treatment conditions rather than between-groups comparisons, and so no studies reached Phase III. That is, no studies used AAN Level II parallel-group designs. Such investigations represent advances relative to the model of clinical outcomes treatment research (Robey & Schultz, 1998) but identify the need to progress further to more advanced experimental designs including systematic group-level randomized controlled trials that test the benefits of specific treatments against each other or against no treatment. It is encouraging, however, to observe that the quality of experimental design, level of participant and treatment descriptions, and the scope of measurement have all been raised.

Of note, 58% of studies (15/26) met all criteria for an AAN Class III rating except for use of independent assessors in scoring outcome measures. To capture the improvement in experimental design, despite lack of independent assessors, we assigned a qualified rating of AAN Level III-b. However, we urge researchers to address this limitation in study design in the future to minimize bias in estimation of treatment effects.

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Table 4. Treatment, maintenance, and generalization outcomes for the 21 studies classified as AAN level III or III-b. Diagnosis rating is repeated here to facilitate interpretation of the outcome data.

First Author (year)	Diagnosis rating	Main approach	Statistical analysis	Treatment outcome		Maintenance[a]		Generalization	
								Response	Stimulus
				Primary measure	Cases with positive outcome	Cases with positive outcome	Tested >2 weeks	Cases with positive outcome	Cases with positive outcome
Articulatory—kinematic approaches									
Austermann Hula (2008)	A	Phonetic placement with 100%/50% feedback frequency or immediate/ delayed feedback	N	Accuracy of words	2/4 on 50% feedback; 1/2 on delayed feedback	Phase I: 3/4; Phase II: 2/2	Yes	Phase I: 3/4 Phase II: 2/2	NR
Ballard (2007)	A	Phonetic placement, variable practice	N	Voice onset time	2/2	2/2	Yes	Yes, 2/2	NR
Cowell (2010)	B	Integral stimulation and imagined production (self-administered)	Y	Accuracy & duration of words	5/6	Not analyzed	Yes	NR	NR
Davis (2009)	A	Implicit phoneme manipulation	Y	Accuracy of phoneme in words	1/1	1/1	Yes	Yes, 1/1	Yes, 1/1
Friedman (2010)	A	Integral stimulation	N	Accuracy of phrases	1/1	1/1	Yes	Yes, 0/1	NR
Katz (2010)	A	Visual kinematic feedback, 100%/ 50% feedback frequency	Y	Accuracy of CV in CVC words	1/1	1/1, superior with 100% feedback	Yes	Yes, 1/1	NR
Kendall (2006)	B	Phonomotor rehabilitation	Y	Accuracy of isolated phonemes	1/1	1/1	No	Yes, 1/1 for 1-syllable words only, ceiling effects	No, 0/1
McNeil (2010)	A	Visual kinematic feedback	Y	Accuracy of phonemes in words	2/2	1/2	Yes	Yes, 2/2	NR
Rose (2006)	A	Verbal ± gestural training	Y	Accuracy of words	1/1	1/1, all conditions	Yes	Yes, 1/1	Yes, 1/1
Savage (2012)	B	Phonetic placement therapy	Y	Accuracy of phonemes in words	1/1	1/1 (anecdotal)	Yes	NR	NR
Schneider (2005)	C	Modified 8-step continuum, high- or low-stimulus complexity	Y	Accuracy of syllable sequences	3/3	NR	No	Yes, 1/3	Yes, 1/3
van der Merwe (2007)	A	Speech motor learning program	N	Accuracy of words and nonwords	1/1	NR	No	NR	NR

(*table continues*)

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Table 4 *(Continued).*

First Author (year)	Diagnosis rating	Main approach	Statistical analysis	Treatment outcome		Maintenance[a]		Generalization	
								Response	Stimulus
				Primary measure	Cases with positive outcome	Cases with positive outcome	Tested >2 weeks	Cases with positive outcome	Cases with positive outcome
van der Merwe (2011)	A	Speech motor learning program	Y	Accuracy of words and nonwords	1/1	1/1	Yes	Yes, 1/1	Yes
Wambaugh (2004)	A	Sound production treatment	N	Accuracy of phonemes in words, phrases, sentences	2/2	Yes, 1/1 tested	Yes	Yes, 2/2	Yes, 2/2
Wambaugh, Nessler (2004)	A	Sound production treatment	N	Accuracy of phonemes in words	1/1	Yes, 1/1	Yes	Yes, 1/1 but limited	Yes, minimal
Wambaugh (2010)	A	Sound production treatment	Y	Accuracy of phonemes in words	1/1	Yes, 1/1	Yes	Yes, 1/1	NR
Wambaugh (2012)	A	Repeated practice ± rate/rhythm control	Y	Accuracy of phonemes in words	8/10	Yes, 8/8	Yes	Yes, 6/10 for frequently probed list, 0/10 for infrequent	NR[b]
Whiteside (2012)	A	Auditory perceptual (input), imagined production, spoken production vs. sham (self-administered)	Y	Accuracy, struggle on word repetition	Superior for speech treatment (n = 44)	Yes, statistically significant	Yes	Yes, not significant	NR
Youmans (2011a, 2001b)	A	Script training	N	% correct production of words	3/3	Yes, 2/2 tested	Yes	NR	NR
Rate and/or rhythm control approaches									
Brendel (2008)	A	Metrical pacing or integral stimulation	Y	Segmental errors, dysfluency, sentence duration	8/10, superior for metrical pacing	Yes, 8/8 tested	Yes	Yes, 8/10	NR
Mauszycki (2008)	A	Metrical pacing	N	Accuracy of words, utterance duration	1/1	Yes, 1/1	Yes	Yes, 1/1	Yes, minimal

Note. NR = not reported.

[a]Considered only testing of long-term maintenance, completed after all treatment(s) had ended. [b]Reported in Wambaugh, Nessler, Mauszycki, and Cameron (2014).

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Improvements in Description of Study Participants

One prerequisite to determining the efficacy of any treatment approach for AOS is the level of confidence that study participants actually demonstrated behaviors consistent with AOS. It was encouraging to observe an increase in the number of studies that permitted reasonable confidence in the diagnosis of AOS. The number of studies receiving an A rating (high to reasonable confidence in diagnosis) has steadily increased from 8% (1/12) in the 1970s to 27% (6/22) in the 1980s, 43% (6/14) in the 1990s, and 69% (18/26) in the 2004 to 2012 period covered herein. Although this surely is a step forward, the rating system we used required the authors merely to list the characteristics that they observed in their participants. In most cases, no evidence was provided in the form of quantitative scores on a diagnostic task or frequency counts of behaviors, and, therefore, evidence supporting authors' severity ratings was not evaluated. Currently, there is no accepted protocol or guideline for assessing speech production and determining when an observed characteristic or combination of characteristics reaches a critical threshold to be considered indicative of AOS or of a specific severity level of AOS, though promising efforts are underway (e.g., Ballard et al., 2014; Haley, Jacks, De Riesthal, Abou-Khalil, & Roth, 2012; Vergis et al., 2014; Whitwell et al., 2013).

Improvements in Description of Treatment

Despite the limitations in diagnostic methods, there is strong consensus that AOS represents impaired speech motor planning and/or programming. As such, it is not surprising that treatments targeting speech motor planning and/or programming, including articulatory–kinematic and rate/rhythm control approaches, have predominated the intervention literature. In the 2006 AOS guidelines report, four general categories of treatment approaches were identified: (a) articulatory–kinematic, (b) rate/rhythm control, (c) intersystemic facilitation/reorganization, and (d) AAC (Wambaugh et al., 2006). Over 90% (24/26) of the studies in the current review article applied an articulatory–kinematic approach; of the remaining two studies, one provided solely rhythm/rate control treatment and one compared these two approaches.

The articulatory–kinematic approaches included a range of techniques including integral stimulation, phonetic placement, visual kinematic feedback on articulator placement and trajectories, adjuvant transcranial direct current stimulation, and combined articulatory plus rhythm/rate or music therapy. In all cases, articulation accuracy was of central interest, with several studies also including measures of speech rate or utterance duration, speech intelligibility, or dysfluency and struggle. Two studies included AAC, using a speech-generating device to provide models for imitation during home practice (Lasker et al., 2008, 2010). This was coded as an articulatory–kinematic study because the focus was on the participant's speech production and the device functioned as a surrogate clinician. One other study might have been coded as using intersystemic reorganization because it compared gesture training with verbal training and gesture plus verbal training in one participant (Rose & Douglas, 2006). Given that the focus was on speech production and the three approaches were considered to be equally efficacious, we grouped this study with other articulatory–kinematic approaches. Currently, we have little or no evidence to support any of these articulatory–kinematic approaches over another because comparative studies have not been done.

Innovations in treatment have also occurred over the past decade of AOS treatment research. It is not surprising that technological advances have been incorporated into the therapeutic process. The first application of transcranial direct current stimulation has been reported with encouraging findings (Marangolo et al., 2011). Self-administered, computerized therapy has also been evaluated for the first time with AOS participants (Cowell et al., 2010; Whiteside et al., 2012) with a positive outcome noted. In addition, a series of investigations of the effects of electromagnetic articulography suggested benefits of augmented visual biofeedback (Katz et al., 2010; McNeil et al., 2010). In contrast, very few applications of technology were evident in the studies included in the 2006 guidelines.

PML (Schmidt, 1975; Schmidt & Bjork, 1992) have been suggested for use in the treatment of AOS for many years (Robin, 1992; Duffy, 1995; McNeil et al., 1997). However, only a few studies included in the first AOS evidence-base review article addressed these principles (e.g., Knock, Ballard, Robin, & Schmidt, 2000; Maas, Robin, Barlow, & Shapiro, 2002). In the current review article, there is obviously a trend to incorporate principles of motor learning in AOS treatment. Several investigations explicitly evaluated specific principles (e.g., Austermann-Hula et al., 2008; Katz et al., 2010; Schneider & Frens, 2005), and many more have included these principles in the structure of their intervention protocol (Ballard et al., 2007; Friedman et al., 2010; Lasker et al., 2008, 2010; Rose & Douglas, 2006; van der Merwe, 2011). Bislick and colleagues (2012) recently reported a systematic review that addressed PML and speech motor learning and concluded that findings were promising, albeit limited at this time. There is little doubt that future studies will continue to explore the effects of these principles on the impaired speech motor system.

It is worth noting two studies here that were not included in the current review article because they considered factors influencing change in speech motor skill over brief time frames and were considered facilitation studies rather than treatment studies (Aichert & Ziegler, 2008; Schoor, Aichert, & Ziegler, 2012). These studies involved participants with AOS and provided intensive practice on syllable production, with measures of production accuracy for trained and related untrained syllables. Both demonstrated convincing transfer of training effects to syllables related by syllabic structure. These studies provide interesting insight into the structure of speech motor plans, indicating that phonemes are linked to syllabic nodes (i.e., onset and

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rhyme) and supporting intervention approaches that target the syllable as the smallest unit of motor planning. Perhaps the next review will see full-scale evaluation of long-term learning with this approach that carefully manipulates syllable structure to maximize practice effects.

Measurement of Treatment Effects

Treatments were typically applied for about 28 sessions over at least 7 weeks. As expected, the most frequently used primary outcome measure was perceptually judged accuracy of phoneme or word production. For the rhythm/rate control treatments, additional measures included word or utterance duration, speech rate, and/or dysfluency. Fourteen of the articulatory–kinematic studies reported using specific PML (Schmidt & Lee, 2011). The principles applied or tested were level of feedback frequency, timing of feedback relative to participant's response, using variable practice (i.e., stimuli varied along some dimension such as voice onset time or phonetic context) and random versus blocked stimulus presentation, and using high-complexity (consonant clusters) versus low-complexity (singletons) stimuli.

One additional principle considered beneficial for motor learning is high-intensity practice, reflected in number of practice trials per sessions and/or number of sessions per week. It was not possible to analyze within-session intensity due to lack of detail in many studies; however, the typical session frequency was moderate at three per week, with a typical intervention duration of 7.5 weeks. Warren, Fey, and Yoder (2007) provided a framework for quantifying treatment intensity, which may guide the design and reporting of studies going forward. Specifically, researchers are encouraged to report on dose (e.g., the number of productions elicited from the participant per session), dose form (e.g., individual or group sessions, controlled or incidental elicitation of target behaviors), dose frequency (i.e., number of sessions per unit of time), total intervention duration in weeks/months, and, finally, cumulative intervention intensity representing Dose × Dose Frequency × Total Intervention Duration. With such information, potentially it will be possible to evaluate whether treatment intensity is a key factor in outcome (see Baker, 2012) and how intensity might interact with different treatment approaches.

Limitations and Future Directions

Important advances stemming from the past decade of AOS treatment research are evident. However, many of the research needs identified in the 2006 report remain. Although significant improvements have occurred relative to replication of treatment effects across participants, replication is still insufficient. For single-subject experimental studies, the minimum standard of three replications to demonstrate experimental control (within-case or across cases) (Kratochwill et al., 2010) has rarely been met. It is important to note there are no replications of any group or single-subject investigations across independent laboratories.

As a whole, the AOS treatment studies in this report suggest a progression from early Phase I investigations to studies reflecting later phases of the clinical outcomes treatment research model. However, there are very few instances in which a given treatment has been systematically advanced through the model. Consequently, as discussed by Wambaugh et al. (2006a, 2006b), many important issues have not been addressed such as optimum treatment intensity/dosage, criteria for discharge, and variations in treatment response related to participant characteristics.

Outcome measures continue to be problematic from several perspectives. As was the situation at the time of the previous systematic review (Wambaugh et al., 2006a, 2006b), there has been little attention given to understanding the effect of treatment beyond impairment level measures. This likely stems from the lack of development or availability of psychometrically sound outcome measures that address speech-related activity limitations, participation restrictions, and psychosocial well-being.

This review has only considered articles published in English, and conclusions may also have been influenced by a lack of published negative findings (Hopewell et al., 2009). It is also possible that the search process missed some eligible studies.

A meta-analysis has not been performed here, but we refer readers to Strom (2008), who completed quantitative meta-analyses of eight group and 11 single-subject design studies reporting outcomes for articulatory–kinematic treatments, grouped together, using measures of correct words or sounds. As stated earlier, Strom concluded that, in general, articulatory–kinematic treatments for AOS are efficacious. This conclusion is consistent with the interpretation of the current review. What we do not yet know is whether some articulatory–kinematic treatments are more efficacious than others and possible reasons for such differences. Future work should include randomized control trials to address this gap.

This report was focused only on studies published since the original 2006 AOS guidelines report. A consideration of the entire AOS evidence base is also warranted to draw conclusions concerning the status of the evidence supporting specific treatments or treatment approaches. In the 2006 report, studies were grouped by general approach because there was a lack of evidence for any one treatment. Given the increase in the AOS evidence base, it appears likely that there are now sufficient numbers of studies directed toward particular treatments/approaches to warrant examination of the evidence in a more fine-grained manner. For example, within the general category of articulatory–kinematic approaches, multiple investigations are available for several treatments (e.g., sound production treatment, eight-step continuum, motor learning guided treatment). The current project was deemed necessary as a step toward determining the need for evaluation of discrete treatments or subcategories of treatments. A

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follow-up project is underway to critically assess the evidence associated with specific treatments.

In the present report and the original AOS guidelines project, the granularity of the judgments of scientific adequacy could be considered rather broad. That is, there are methods that may be considered more fine-grained (and perhaps more rigorous, particularly for single-case experimental designs) than those used (e.g., Kratochwill et al., 2010). For the follow-up project, rigorous standards are being used to evaluate the experimental methods of each study contributing the evidence for a particular treatment. It is anticipated that a relatively small subset of the 80+ studies that constitute the entire AOS evidence base will be evaluated; thus, a more refined level of analysis will be possible.

Conclusions

Continued research over the 9 years from 2004 to 2012 has augmented the treatment database. We find it encouraging that as a group, these recent studies have improved in diagnostic specification and rigor of experimental design. The most important global clinical conclusion supported by this review is that the weight of evidence supports a strong effect for both articulatory–kinematic and rate/rhythm-based interventions (see also Strom, 2008). Many studies represent fruitful directions for establishing high-level evidence for theoretically grounded and well-designed intervention for AOS and are promising candidates for larger scale studies. It will be through such large-scale studies, including treatment comparisons, that an understanding of the interactions between patient variables and treatment conditions will be developed. This understanding will provide the bases for predicting which treatments will be most appropriate at a given severity level, for a given behavior type or communication context, or for the stage of recovery or progression.

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