

Treatment Guidelines for Apraxia of Speech: Lessons for Future Research

Julie L. Wambaugh, Ph.D.

*VA Salt Lake City Healthcare System and University of Utah
Salt Lake City, Utah*

The Academy of Neurologic Communication Disorders and Sciences (ANCDS) Writing Committee of Treatment Guidelines for Apraxia of Speech (AOS) prepared a technical report containing evidence-based guidelines for the management of AOS. The technical report, which was based on a comprehensive review of published treatment literature, revealed numerous limitations in that literature. This report utilizes information gleaned from the AOS Treatment Guidelines to provide suggestions for future research focused on the treatment of AOS. Ideas are offered for strengthening the methods employed in AOS treatment investigations and for possible directions of research.

The Academy of Neurologic Communication Disorders and Sciences (ANCDS) appointed a writing committee that was asked to develop evidence-based treatment guidelines for acquired apraxia of speech (AOS). The AOS treatment guidelines project, initiated in 2001, is part of the larger endeavor of ANCDS to develop practice guidelines for various neurologically impaired patient populations (Frattali et al., 2003; Golper et al., 2001).

The AOS Committee systematically reviewed all of the existing English-language publications pertinent to the treatment of AOS in the process of developing guidelines. A summary of that evidence, along with the corresponding treatment guidelines, has been prepared as a technical report and clinically oriented reports (Wambaugh, Duffy, McNeil, Robin, & Rogers, 2006a, 2006b, 2006c).

An important part of guideline development requires that the developers provide suggestions for future research, so that the treatment evidence base may be ultimately strengthened (Frattali et al., 2003). Although the AOS Committee identified general areas requiring additional or improved research (Wambaugh et al., 2006c), specific suggestions have not yet been provided. Therefore, the purpose of

this report is to provide relatively explicit suggestions regarding AOS treatment research, which were derived from the development of the AOS treatment guidelines.

The initial portion of this report will focus on the methods employed in AOS treatment research, and the remaining sections will concentrate on directions for future research.

SUGGESTIONS FOR IMPROVING METHODS

Participant Diagnosis and Description

The AOS Treatment Guidelines report indicated that a particularly problematic area in the AOS treatment literature was the description of the participants in the investigations. Across the majority of the investigations reviewed by the AOS Committee, there was insufficient information provided concerning AOS diagnosis, presenting speech behaviors, severity levels, and cooccurring speech/language disorders (Wambaugh et al., 2006c).

Given the fact that at the current time there continues to be lack of agreement on diagnostic stan-

dards for AOS, it is imperative that investigators cite the specific criteria used in determining the diagnosis of AOS (e.g., McNeil, Robin, & Schmidt, 1997). This specification should be accompanied by a description of the conditions under which the diagnosis is made. That is, the person(s) making the diagnosis should be well-described in terms of degree of experience, familiarity with the participant(s), awareness of diagnostic possibilities (e.g., blinded to previous diagnosis and purpose of investigation), and level of independence during diagnosis (e.g., single, independent judgment vs. agreement of multiple, independent examiners vs. consensus of multiple, nonindependent examiners).

Additionally, the speech samples utilized to make the diagnosis should be described. For example, the investigator should indicate if the diagnosis was based on a sample from a single session or repeated samples and whether the samples were live and/or recorded. If a test (e.g., Dabul, 2000) or published assessment protocol (e.g., Duffy, 2005) was not employed and cited, the type of speech sample(s) utilized in the diagnostic process should be explained (e.g., repeated productions of multisyllabic words, AMRs and SMRs, narrative discourse sample, oral reading of short sentences, etc.).

Beyond describing the diagnostic process, the investigators should characterize the behaviors that support the diagnosis with enough detail to allow an estimation of the level of severity of the AOS. At a minimum, this should depict the level of production at which AOS behaviors occurred and the types of errors that were present (Example 1). Preferably, the investigator should provide examples or summaries of sound errors and other AOS behaviors (Example 2: Table 1).

Example 1

P 1 evidenced no sound errors when producing monosyllabic and bisyllabic words in a repetition task. During repetition of words of three syllables or greater, he produced sound errors on approximately 50% of the words. Those errors tended to be distortions and occurred primarily on fricatives. In oral reading of sentences and production of narrative discourse, P 1 produced errors on approximately 25% of attempted sounds. Rate was noticeably reduced in all contexts other than monosyllabic word production, with syllable segregation evident on the majority of multisyllabic word productions.

The AOS Committee documented the provision of various descriptive variables thought to be pertinent to treatment investigations (Wambaugh et al., 2006a). Although "language information" was provided in most investigations, minimal information concerning the impact of the cooccurring aphasia on verbal production was typically available. Speech and language disorders that co occur with AOS are likely to influence response to treatment and should be carefully documented. Minimally, the type and severity of the disorder(s) and effects on production should be described (Example 3).

Example 3

Participant 1 presented with mild, anomic aphasia (PICA -85th %tile; WAB-classification), which had reportedly resolved from moderate, nonfluent aphasia. His word retrieval difficulties resulted in increased latencies during confrontation naming and infrequent phonemic paraphasias during confrontation naming and spontaneous speech. No dysarthria, as described by Duffy (2005) was noted.

Several other variables were reported in a relatively low percentage of the studies reviewed by the

TABLE 1. Example 2: Percent occurrence of sound errors and AOS behaviors across speaking contexts.

Speech Sample	Sounds in Error	Distort.	Voicing Errors	Omis.	Subst.	Schwa Additions	Syll. Seg. or ↑ iwi's	↓ Rate
Monosyllabic	0						na	0
BiSyllabic	0						50	50
TriSyllabic	25	20	70	0	10	0	100	100
4+ Syllables	40	25	60	0	10	5	100	100
Sentences*	30	25	70	0	5	0	25	50
Discourse **	30	20	70	0	5	5	30	75

* repetition of sentence of 6–8 syllables comprised of monosyllabic and bisyllabic words

** 2 minute narrative discourse sample (cite conditions)

Distort. = distortions not including voicing errors, Subst. = perceived substitutions

Omis. = omissions, Syll. Seg. = syllable segregation, iwi = interword intervals

AOS Committee: race/ethnicity, socio-economic status and education, cognitive functioning, hearing, medications, and neuroimaging information (Wambaugh et al., 2006a). Currently, the impact of these variables on response to treatment is unknown. However, routine provision of this information may allow inferences regarding prognosis to be made when a substantial body of data is available.

Internal and External Validity

Approximately two-thirds of the investigations that comprised the evidence base for the AOS Treatment Guidelines lacked the scientific control necessary to assure that treatment was responsible for observed changes (Wambaugh et al., 2006b). This was not surprising, given that more than half of the investigations were case studies. Although a trend of increasing utilization of single-subject experimental designs was observed (Wambaugh et al., 2006b), it is imperative for the advancement of AOS treatment that researchers employ experimental designs whenever possible.

Although Randomized Controlled Clinical Trials (RCTs) are considered the gold standard of evidence, such trials are probably not appropriate at this point in time for the majority of AOS treatments. Basic issues relative to treatment efficacy have not been clarified for most treatment approaches (see below). Furthermore, RCTs may never be feasible for AOS treatments due to limiting factors such as the requirement of relatively large number of subjects, the heterogeneity of the population, and the confound of cooccurring aphasia. At the current time, relatively small "n" group studies, single-subject designs, and combined group–single-subject designs appear to be the most appropriate and/or viable options for AOS treatment research.

As noted previously, single-subject designs have been employed in increasing proportions in AOS research and information from past design applications may be utilized to strengthen the design and implementation of future AOS treatment research efforts.

To enhance internal validity, multiple baseline designs (MBD) **across subjects** should always be considered as a design component if more than one participant is being treated, regardless of type of overall single-subject design (e.g., ABA, multiple baseline across behaviors, alternating treatments design). By extending baselines and replicating treatment effects across participants, the MBD-across subjects design provides additional experimental control and is especially important in cases

where little is known about generalization effects of treatment (see below).

MBDs-across behaviors have been the most commonly employed single-subject design with AOS (Wambaugh et al., 2006a). Such designs may or may not be appropriate for all AOS treatment approaches because of the requirement of independence of the multiple behaviors (Barlow & Hersen, 1984). On the basis of the extant literature, it appears that response classes are limited in articulatory-kinematic (A-K) treatments of AOS (Wambaugh et al., 2006c). That is, generalization **across** sounds is not expected unless sound errors and sound targets are closely related (i.e., behaviors are largely independent). Consequently, MBD-across behaviors designs are appropriate for studies of A-K treatments unless treatment is applied to all sounds in error simultaneously. Previous research has indicated that for A-K treatments, **within** sound generalization (i.e., to untrained exemplars of trained sounds) is expected, so it would not be appropriate to establish untreated exemplars as a control behavior.

For other treatments, such as rate control and intersystemic reorganization, response classes have not been clearly established. Therefore, unexpected generalization may occur to untrained behaviors, resulting in loss of experimental control. When generalization effects cannot be predicted, it is strongly recommended that a MBD-across subjects design be incorporated if a MBD-across behaviors is utilized.

Beyond concerns over independent behaviors, there are other issues concerning MBDs and AOS treatments that should be considered. The extended probing required with MBDs may have undesired effects in that repeated exposure (1) may result in improved responding of untrained behaviors and result in a loss of experimental control (e.g., Wambaugh & Nessler, 2004) or (2) may be detrimental to treatment because behaviors are repeatedly elicited and not penalized if incorrect, thus potentially reinforcing the incorrect behavior.

The multiple probe design (MPD; Horner & Baer, 1978) is a variant of the multiple baseline design that has been utilized infrequently in AOS treatment studies, but appears to be promising. The MPD provides experimental control similar to a MBD, but employs a reduced probing schedule, which may reduce concerns over the implementation of single-subject designs in a clinical setting. Furthermore, MPDs may alleviate the potential problems associated with excessive probing in MBDs.

An extremely strong single-subject design that has been used infrequently in the AOS treatment

literature, but has potential for wider application is the withdrawal design (e.g., ABAB). As seen in the evidence table developed as part of the AOS guidelines project (Wambaugh et al., 2006a), withdrawal of treatment in multiple baseline designs often resulted in a decrease in performance of trained behaviors. Like other motor skills, it appears that permanent changes in speech behaviors likely require a degree of overlearning (i.e., repeated practice at high levels of accuracy over time). Withdrawal designs require behavior to “reverse” toward baseline levels on removal of treatment. On the basis of the existing literature, it is probable that withdrawal effects will be present if treatment is withdrawn **very soon after improvements have been observed**. This would require the investigator to predetermine the level at which treatment effects would be clear (based on baseline performance and desired level of clinical and/or statistical significance and if possible, predicted level of A prime performance) and withdraw treatment immediately on that criterion being reached.

Group investigations have been used so infrequently in AOS treatment research that no lessons from such applications are available. Preexperimental (e.g., one group, prepost test design), quasi experimental (e.g., time-series, equivalent control group, pretest–posttest nonequivalent designs), and experimental designs (e.g., pretest–posttest randomized control group design) would all be appropriate considerations for future AOS treatment research.

The lack of replication of treatment effects within and across AOS investigations is a major weakness of the AOS treatment evidence base (Wambaugh et al., 2006c). For single-subject designs, a minimum of three replications across subjects is considered necessary for the demonstration of treatment “efficacy” (Chambless & Hollon, 1998). Such replications may be cumulative (i.e., across time or studies). Obviously, treatment investigations involving a single participant are labor intensive and findings are of value. However, it is imperative from both research and clinical perspectives that positive findings be replicated. Furthermore, for both group and single-subject designs, replication of findings across independent laboratories is desired (Chambless & Hollon, 1998).

SUGGESTIONS FOR FUTURE RESEARCH DIRECTIONS

The AOS Guidelines Committee identified four general categories of AOS treatments: (1) articulatory

kinematic (AK), (2) rate and/or rhythm, (3) alternative/augmentative communication (AAC), and (4) intersystemic facilitation/reorganization. The majority of the AOS treatment literature available involved AK treatments (52% of the studies), with few studies devoted to the other types of treatment (Wambaugh et al., 2006a). Even in the area of AK AOS treatment, the available evidence represents the earliest phases of treatment development and testing (after Robey & Schultz, 1998). That is, research has been largely devoted to detecting the activity of a treatment and developing hypotheses for future research.

In keeping with Robey and Schultz’s (1998) model of clinical outcome research, the logical extension of research involving AK treatments would involve experiments designed to test variations in treatment protocols and treatment participants. Perhaps one of the most important protocol issues to be addressed in future studies is that of dosage; specifically, the schedule and amount of treatment for maximizing positive outcomes should be determined. It is likely that dosage will be dependent to an extent on participant characteristics. Most of the research concerning AK treatments has involved participants judged to have severe AOS (84%; Wambaugh et al., 2006c). Systematic replications across participants of varying severities and AOS presentations are warranted for all existing AOS treatments. AK treatment investigations have typically investigated the effects of therapies comprised of numerous techniques (i.e., treatment packages), with little attention paid to the effects of the individual therapy components (except see Simmons, 1980). Recent work focused on variations in feedback delivery (Austerman Hula, Robin, Maas, Ballard, & Schmidt, 2006) illustrate the type of protocol testing necessary to advance this area of study.

The areas of rate and rhythm control, intersystemic facilitation/reorganization, and AAC have received relatively limited study and consequently, additional investigations documenting the presence of therapeutic activity are needed. Following clear documentation of behavioral changes with treatment, additional research as described above would be appropriate.

For all AOS treatments, there is a need for development of outcome measures beyond speech intelligibility and treatment-specific probes. Valid and reliable measures that document changes in speech production at the level of spontaneous or elicited discourse are not available. Also lacking are tools to assess the impact of treatment on activity limitations and participation restrictions. The cur-

rent unavailability of such measures should not prohibit investigators from attempting to estimate the effects of treatment at such levels; however, caveats regarding reliability should be provided when interpreting findings.

Given the immature state of AOS treatment research, it is not unexpected that there is almost no research comparing treatments or examining the effects of combined treatment approaches. However, as AK treatments have been determined to be "probably effective" (Wambaugh et al., 2006c), comparisons of AK treatments that have been shown to have clear effects would be beneficial. Similarly, the incorporation of other treatment approaches with AK approaches (e.g., rate control combined with AK treatment; intersystemic facilitation/ reorganization combined with AK treatment) may have potential for investigation.

Researchers have ample opportunities for advancing this area of inquiry and clinical practice.

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Address correspondence to Julie Wambaugh, 151 A, 500 Foothill Boulevard, Salt Lake City, UT 84148 USA.

e-mail: julie.wambaugh@health.utah.edu

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