

# Effectiveness of Interventions for Preschool Children with Fluency Disorders: A Comparison of Direct Versus Indirect Treatments

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## Introduction

The Individuals with Disabilities Education Act (Public Law 102-119) has shed light on the need and benefits of early intervention for children with speech and language disorders, including stuttering. Stuttering research suggests that children under 3 years of age are at greatest risk of onset (Yairi & Amborse, 1992), with fluency more amendable to improve in the early stages (Adams, 1984; Gottwald & Starkweather, 1995; Onslow, 1992; Starkweather, Gottwald & Halfond, 1990). As such, speechlanguage pathologists (SLPs) play a vital role in the early management of these children. SLPs provide treatment in both home and school settings, with the overall goal of improving functional communication and eliminating dysfluencies. For preschoolaged children between 3 and 5 years of age, clinicians employ a number of direct and indirect behavioral treatments that focus on (a) teaching the child specific ways to modify and correct dysfluent speech patterns and/or (b) working with caregivers/parents to facilitate fluent speech by manipulating the child's environment. A recent systematic review by Bothe, Davidow, Bramlett, & Ingham (2006) explored the efficacy research for behavioral as well as other cognitive and related treatments from 1970 to 2005. This review examined the evidence across all age spans and included nine studies with participants  $\leq 6$  years of age. The majority of these studies (8/9) investigated the use of response-contingency treatments (direct treatments), and only one study (Franken, Kielstra-Van der Schalk & Boelens, 2005) evaluated the comparative effects of a response-contingency direct treatment (i.e., the Lidcombe Program; Onslow, Packman, & Harrison, 2003) and an indirect treatment based on the demands and capacities model (DCM; Starkweather, Gottwald, & Halfond, 1990). The remaining study examined the effects of language training (Butcher, McFadden, Quinn, & Ryan, 2003). Overall findings of the Bothe review for this age group revealed positive outcomes for direct treatments using response-contingency principles. The authors indicated that preschool children receiving direct treatment "at least show arguably better progress than children not exposed to treatment, when data from a control group are reported" (p. 334). No differences were noted in the Franken et al. (2005) study, with an average reduction in percent of syllables stuttered for children in both indirect and direct treatment groups. An additional review that included preschooland school- aged children by Herder, Howard, Nye, & Vanryckeghem (2005) found similar results for behaviorally based interventions.

The results of both reviews are an essential step in the clinical decision-making process and highlight behavioral treatments that have been tested and shown to be effective for preschool children with fluency disorders. These findings—in combination with clinical expertise and client/family preferences—will assist SLPs in making treatment decisions. Moreover, the findings illuminate areas of stuttering research that are in need of further investigation. In an attempt to expand on these early works, the following text further explores the comparative effects of direct and indirect treatments.

#### Method

Six clinical questions were established a priori for review to allow examination of the immediate and long-term effects of direct versus indirect fluency treatments for preschool-aged populations (see Appendix A1). Criteria for considering studies under review are outlined below.

## **Types of Studies**

The types of studies that were examined included all experimental or quasiexperimental studies comparing two types of interventions—direct and indirect treatments. Individual studies that were included in an accepted systematic review or meta-analysis were excluded.

# **Types of Participants**

Participants were children ages 3-5 years who have received the diagnosis of a fluency disorder (group Ms < 6 years of age; participants must not be in kindergarten). The diagnosis of a fluency disorder could be the primary diagnosis or could be secondary to another condition. Studies including mixed ages or populations were excluded unless data could be separated for analyses.

# Types of Interventions

Studies examining any indirect fluency treatment approach compared with a direct fluency treatment were included. *Indirect treatments* were operationally defined as any treatment modifying the communication partner's speech and behavior to improve the child's fluency. Any treatment with the primary focus on improving fluency with minimal involvement of the child was considered *indirect*. Examples include caregiver changing his/her interaction style by slowing down, using simple vocabulary, not criticizing or pressuring child, using parent modeling techniques, and applying manualized treatment approaches such as parent–child interaction therapy (Egolf, Shames, Johnson, & Kasprisin-Burrelli, 1972) and the demands and capacities model. *Direct treatments* were operationally defined as any treatment focused on manipulating the child's communication to improve fluency. Direct treatments require the child to make specific

changes to his/her speech production. Examples include, but are not limited to, treatments that focus on slowing speech rate, prolonging vowels, teaching soft consonant or easy onset techniques, phrasing and pausing techniques, modeling, fluency shaping, and auditory feedback training. Manualized treatment approaches also include the Lidcombe Program, the Fluency Development System for Young Children (Meyers & Woodford, 1992), and Gradual Increase in Length and Complexity of Utterance (GILCU; Ryan, 1974). Studies that use an alternative treatment approach with a separate approach for each treatment phase were included. Studies that utilize both treatment approaches (e.g., Fun with Fluency; Walton, Wallace, & Anderson, 1998) were excluded unless data could be separated for analyses. Studies were excluded if they employed pharmacological interventions or utilized animal models.

#### **Types of Outcomes**

Studies reporting pre- and post data for one or more *speech*, *social/emotional* or *parent* outcome were explored. *Speech outcomes* included, but were not limited to, percentage of dysfluencies per syllable, percentage of dysfluencies per word, speaking rate, and so forth. *Social/emotional outcomes* included any measures of the child's communication attitude or behavior (e.g., communication attitude test, severity rating scales, self-report rating, quality-of-life indicators, social communication measures, etc.). *Parent outcomes* included any measure of the parent's attitude toward the child's communication (e.g., parent rating scales, parent surveys) or change in parent's communication behavior (e.g., pre/post parent communication patterns, etc.). In addition, short-term and long-term effects of treatment were considered. *Short-term outcomes* were operationally defined as immediate post-treatment outcomes, and *long-term outcomes* were defined as follow-up or maintenance of treatment outcomes.

#### Search Methods for Study Inclusion

English-language, peer-reviewed studies were identified through the search of 22 electronic databases. Given that two previous reviews (Bothe et al., 2006; Herder et al., 2005) examined the scientific research pertaining to stuttering interventions that included direct and indirect treatments, a search date of "2005 to present" was selected

to identify any new peer-reviewed literature. See Appendix A2 for a full list of search parameters, search strategy, and databases. A manual search of all relevant references and prolific authors was also conducted.

## Study Appraisal and Data Extraction

Accepted studies were independently evaluated for methodological quality by two coders on eight quality indicators:

- Adequate description of protocol for replication
- Adequate description of subjects (within-subject design) or groups comparable at baseline (between-subject design)
- Assessors blinded
- Random sample adequately described
- Evidence of treatment fidelity
- Report of *p* value or calculable from data
- Report of effect size and confidence interval or calculable from data
- Use of intention-to-treat analysis (controlled trials)

Reliability of appraisal ratings between coders was assessed, and any disagreement in ratings was documented and resolved via consensus. Pertinent demographic, intervention, outcomes, and major findings data were extracted from each included study.

# Results

Sixty-two abstracts were identified and independently reviewed by two authors. Level of agreement between authors for study inclusion was 94%. Of the abstracts reviewed, four met preliminary inclusion criteria. However, upon review of full-text citations, two abstracts were further eliminated due to inability to separate data specific to the population under review. An additional study (Medical and Health Research Council of

the Netherlands, 2007) was excluded due to inability to obtain published data<sup>1</sup>. The final study (Franken et al., 2005) was eliminated, as it was included in previous reviews (Bothe et al., 2006; Herder et al., 2005), thus leaving no new studies found.

Reasons for exclusion of studies were as follows: (a) it was not a study, systematic review, or meta-analysis targeting one or more a clinical question (53/62); (b) no original data were available, or original data could not be obtained (4/62); (c) not age or population under review (3/62); or (d) it was not peer reviewed, or it was published in a language other than English (2/62). See Appendix A3 for full bibliography of studies.

## Discussion

Currently, there is insufficient evidence to support or refute the use of a direct intervention approach over an indirect approach in the treatment of preschool-aged children with fluency disorders. At this time, the state of the evidence comparing direct and indirect treatments is limited to one study (Franken et al., 2005). In this study, the authors compared speech and parent outcomes of children randomly assigned to the Lidcome Program (a direct treatment approach) with the Demands and Capacities model (an indirect approach), with no differences in stuttering frequency and severity ratings found.

Unfortunately, the current state of the evidence does not provide meaningful information for clinicians attempting to decide between the use of direct or indirect treatments for stuttering in young children. While considerably more evidence is available in favor of direct treatment approaches, such as the Lidcombe Program, very limited evidence (from one study) also suggests potential success for children treated with indirect approaches such as the Demands and Capacities model. Clearly, more high quality experimental or quasi-experimental studies of well-defined interventions targeted to preschool children with fluency disorders are needed, especially studies comparing the relative effectiveness of one treatment over another.

<sup>&</sup>lt;sup>1</sup> Study abstract appeared to meet inclusion criteria. Author was contacted March 3, 2010, to obtain full report. As per author communication, final report had not been completed and published to date.

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Appendix A1 – Clinical questions under review.

- 1. What is the short-term effect of indirect versus direct treatment on speech outcomes for children ages 3 to 5 with a fluency disorder?
- 2. What is the long-term effect of indirect versus direct treatment on speech outcomes for children ages 3 to 5 with a fluency disorder?
- 3. What is the short-term effect of indirect versus direct treatment on social/emotional outcomes for children ages 3 to 5 with a fluency disorder?
- 4. What is the long-term effect of indirect versus direct treatment on social/emotional outcomes for children ages 3 to 5 with a fluency disorder?
- 5. What is the short-term effect of indirect versus direct treatment on parent outcomes for children ages 3 to 5 with a fluency disorder?
- 6. What is the long-term effect of indirect versus direct treatment on parent outcomes for children ages 3 to 5 with a fluency disorder?

Appendix A2 – Fluency EBSR search methodology.

# **Databases Searched**

- PubMed (http://www.ncbi.nlm.nih.gov/pubmed/)
- CINAHL (EBSCO)
- Health Source: Nursing/Academic Edition (EBSCO)
- Communication & Mass Media Complete (EBSCO)
- Psychology and Behavioral Sciences Collection (EBSCO)
- Education Research Complete (EBSCO)
- ComDisDome (CSA)
- LLBA (CSA)
- CSA Neurosciences Abstracts
- ERIC (CSA)
- CSA Social Services Abstracts
- ScienceDirect
- ISI Web of Knowledge
- Cochrane Library (Wiley)
- SUMSearch (<u>http://sumsearch.uthscsa.edu/</u>)
- speechBITE (<u>http://www.speechbite.com/</u>)
- TRIPDatabase (<u>http://www.tripdatabase.com/</u>)
- Latin American and Caribbean Center on Health Sciences Information (LILACS) (<u>http://www.bireme.br/php/index.php?lang=en</u>)
- Centre for Reviews and Dissemination (http://www.york.ac.uk/inst/crd/)
- HighWire Press
- Health Information Resources, formerly National Library for Health (<u>http://www.library.nhs.uk/Default.aspx</u>)
- GoogleScholar (<u>http://scholar.google.com</u>)

## Search Criteria

- Date of publication 2005 to present (including articles in press).
- English language only.
- Published in a peer-reviewed journal.
- Must be a study with original data that addresses one or more of the clinical question.

## Key Words

- stuttering
- fluency disorders
- speech therapy
- preschool children
- treatment outcome

# Expanded Key Words

("Stuttering"[Mesh] OR "Stuttering/rehabilitation"[Mesh] OR "Stuttering/therapy"[Mesh]) AND ("Speech Therapy"[Mesh])

("Stuttering"[Mesh]) AND ("Behavior Therapy"[Mesh] OR "Cognitive Therapy"[Mesh] OR "Language Therapy"[Mesh] AND "Electromyography"[Mesh])

"Stuttering"[Mesh] AND ("Communication"[Mesh] OR "Speech"[Mesh] OR "Speech Intelligibility"[Mesh] OR "Speech Perception"[Mesh] OR "Phonation"[Mesh] OR "Verbal Behavior"[Mesh]) AND ("Stuttering/rehabilitation"[Mesh] OR "Stuttering/therapy"[Mesh] OR "Speech Production Measurement"[Mesh] OR "Program Evaluation"[Mesh] OR "Parent-Child Relations"[Mesh] OR "Parenting"[Mesh] OR "Time Factors"[Mesh] OR "Treatment Outcome"[Mesh])

("Stuttering/rehabilitation"[Mesh] OR "Stuttering/therapy"[Mesh])

"Stuttering/therapy"[Mesh] AND ("Communication"[Mesh] OR "Speech"[Mesh] OR "Speech Intelligibility"[Mesh] OR "Speech Perception"[Mesh] OR "Phonation"[Mesh] OR "Verbal Behavior"[Mesh] OR "Speech Production Measurement"[Mesh] OR "Program Evaluation"[Mesh] OR "Parent-Child Relations"[Mesh] OR "Parenting"[Mesh] OR "Time Factors"[Mesh] OR "Treatment Outcome"[Mesh] OR "Behavior Therapy"[Mesh] OR "Cognitive Therapy"[Mesh] OR "Language Therapy"[Mesh] OR "Electromyography"[Mesh])

("Stuttering"[Mesh] OR "Stuttering/rehabilitation"[Mesh] OR "Stuttering/therapy"[Mesh]) AND ("Speech Therapy"[Mesh])

("Stuttering"[Mesh]) AND ("Behavior Therapy"[Mesh] OR "Cognitive Therapy"[Mesh] OR "Language Therapy"[Mesh] AND "Electromyography"[Mesh])

"Stuttering"[Mesh] AND ("Communication"[Mesh] OR "Speech"[Mesh] OR "Speech Intelligibility"[Mesh] OR "Speech Perception"[Mesh] OR "Phonation"[Mesh] OR "Verbal Behavior"[Mesh]) AND ("Stuttering/rehabilitation"[Mesh] OR "Stuttering/therapy"[Mesh] OR "Speech Production Measurement"[Mesh] OR "Program Evaluation"[Mesh] OR "Parent-Child Relations"[Mesh] OR "Parenting"[Mesh] OR "Time Factors"[Mesh] OR "Treatment Outcome"[Mesh])

("Stuttering/rehabilitation"[Mesh] OR "Stuttering/therapy"[Mesh])

"Stuttering/therapy"[Mesh] AND ("Communication"[Mesh] OR "Speech"[Mesh] OR "Speech Intelligibility"[Mesh] OR "Speech Perception"[Mesh] OR "Phonation"[Mesh] OR "Verbal Behavior"[Mesh] OR "Speech Production Measurement"[Mesh] OR "Program Evaluation"[Mesh] OR "Parent-Child Relations"[Mesh] OR "Parenting"[Mesh] OR "Time Factors"[Mesh] OR "Treatment Outcome"[Mesh] OR "Behavior Therapy"[Mesh] OR "Cognitive Therapy"[Mesh] OR "Language Therapy"[Mesh] OR "Electromyography"[Mesh]) (stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent) AND (treatment OR therapy OR direct OR indirect)

("2005"[Publication Date] : "3000"[Publication Date]) AND ("Speech Therapy"[Mesh] AND (stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent))

("2005"[Publication Date] : "3000"[Publication Date]) AND ("Speech Therapy"[Mesh] AND (clutter\* OR dysfluent OR dysfluenc\* OR stammer\*))

(MH "Fluency Disorders/TH")

(MH "Fluency Disorders") AND (MH "Speech Therapy+")

(MH "Fluency Disorders")

(MH "Speech Therapy+") AND (stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*)

(MM "Fluency Disorders")

(stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*)

(XX "stuttering")

(stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*)

DE "STUTTERING in children"

(DE "SPEECH therapy" OR DE "GROUP speech therapy") AND (stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*)

(treatment OR therapy OR program OR rehabilitation OR intervention) AND (stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*) AND DE "PRESCHOOL children")

(DE "FLUENCY (LANGUAGE LEARNING)" AND PT "ACADEMIC JOURNAL") OR (DE "FLUENCY (LANGUAGE LEARNING) --\*" AND PT "ACADEMIC JOURNAL")

(stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*) AND (treatment OR therapy OR program OR rehabilitation OR intervention)

DE "Stuttering"

DE "Speech Therapy" AND (stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*)

(stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*)

("stuttering" or "fluency disorders" or "disfluency" or "dysfluency" or "stammering" or "secondary stuttering behaviors" or "accessory stuttering behaviors") and ("speech therapy" or "stuttering intervention") and ("preschool children")

(DE="stuttering 84850" and DE="preschool children 67350" and DE="speech therapy 83200")

(stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*) and ("preschool children")

DE="children" and (stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*)

(DE=("stuttering" and "speech therapy")) and (DE="preschool children")

(DE="preschool children") and (stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*)

Topic=((stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*)) Refined by: Publication Years=( 2009 OR 2008 OR 2007 OR 2006 OR 2005 ) AND Document Type=( ARTICLE OR REVIEW ) AND Languages=( ENGLISH ) AND Topic=(child\* OR preschool\*)

(pub-date > 2004 and TITLE-ABSTR-KEY (stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*) and ALL(treatment OR therapy OR program OR rehabilitation OR intervention OR exercise OR remediation OR therapies)) and child\* OR preschool\* OR pre-school\*

(child\* OR preschool\* OR pre-school\*) AND (stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\*)

(child\* OR preschool\* OR pre-school\*) AND (stammer\*)

(child\*) (stutter\* or OR or fluenc\* or OR or fluent or OR or disfluenc\* or OR or disfluent or OR or clutter\* or OR or dysfluent or OR or dysfluenc\*) from:2005 to:2010 (preschool\*) (stutter\* or OR or fluenc\* or OR or fluent or OR or disfluenc\* or OR or disfluent or OR or clutter\* or OR or dysfluent or OR or dysfluenc\*) from:2005 to:2010

Search for: STUTTER\* OR FLUENC\* OR FLUENT OR DISFLUENC\* OR DISFLUENT OR CLUTTER\* OR DYSFLUENT OR DYSFLUENC\* (Focus: TREATMENT, ages: child, subjects: HUMAN)

Search results for Target area: Fluency (All)

Search on: [MH]"Stuttering"

stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\* RESTRICT YR 2005 2010

Searched: (stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\*) AND (treatment OR therapy OR program OR rehabilitation OR intervention)

"stutter\* OR fluenc\* OR fluent OR disfluenc\* OR disfluent OR clutter\* OR dysfluent OR dysfluenc\* OR stammer\* and treatment OR therapy OR program OR rehabilitation OR intervention OR exercise OR remediation OR therapies

(stutter\* OR fluency OR fluent OR disfluent OR disfluenc\* OR dysfluent OR dysfluenc\* OR clammer\* OR clutter\* OR stammer\*) AND (treatment OR therapy OR therapies OR rehabilitation OR program OR intervention OR exercise OR remediation) AND (preschool\* OR pre-school\* OR child\*) **Appendix A3** – Bibliography of excluded citations.

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