

Language-based interventions for improving linguistic outcomes in children with developmental disorders

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Date Submitted: 7 November 2016

Date Revision Submitted: 23 January 2017

Approval Date:

Publication Date: 24 April 2017

TITLE OF THE REVIEW

Language-Based Interventions for Improving Linguistic Outcomes in Children with Developmental Disorders: A Systematic Review

BACKGROUND

Language is a complex, formal system and poses as a pivotal skill to master in a child's development. Both theory and research highlight the importance of developing language and how this may influence developmental pathways both on a short term and long term basis (Stothard, Snowling, Bishop, Chipchase, & Kaplan, 1998). Language delays or deficits are frequently encountered at child development clinics (O'Hare, 2013) regardless of what diagnostic category the child may belong to. Children belonging to diagnostic groups where problems related to language development are central are often in need of interventions that target language skills. One broad group eligible for such interventions belong under the umbrella terms of developmental disorders or neurodevelopmental disorders (Bishop & Rutter, 2008; D'Souza & Karmiloff-Smith, 2017). Developmental disorders can be grouped in two where one group such as children with Language Disorder (LD) and Autism Spectrum Disorder (ASD) have a presumed, not fully known, multifactorial aetiology (Thapar & Rutter, 2015). The other group have a known genetic or acquired aetiology (e.g. Down Syndrome and Fragile X). Historically, developmental disorders have been associated with intellectual disabilities and cognitive levels below population average. However, the trend, for instance in DSM-5 sees the role of cognitive levels as measured by traditional IQ-tests being downplayed, which in turn make way for focus on adaptive functioning. Regardless, cognitive functioning will remain closely intertwined with developmental disorders and pose as a key factor in influencing possible intervention outcomes (Rice, 2016).

The value of cross-disordered samples

Many studies compare children from different diagnostic groups on various language constructs to investigate uniqueness and similarities. One study of children with Williams Syndrome and children with specific language impairments reported distinct patterns of syntactic binding (Ring & Clahsen, 2005). Differences in language has also been reported between children with Fragile X syndrome and Down Syndrome where also level of autism symptom severity were indicative of differences between groups (Martin, Losh, Estigarribi, Sideris, & Roberts, 2013; Price, Roberts, Vandergrift, & Martin, 2007). On the other hand, children with ASD, DS, WS, ID, or FXS all, to a varying extent, display some kind of delay or deficit of language (Abbeduto, McDuffie, Thurman, & Kover, 2016; Marcus & Rabagliati, 2006; Rice, Warren, & Betz, 2005). Further, other studies of children with developmental disorders report that there is considerable overlap in symptomatology (Gibson, Adams, Lockton, & Green, 2013) and aetiological similarities (Valenti, Bari, Filippis, Henrion-Caude, & Vacca, 2014) as well as commonalities in cognitive development (Raitano Lee, Maiman, & Godfrey, 2016). Additionally, there is high comorbidity between these groups of children

(Abbeduto et al., 2016; APA, 2013) and the different diagnoses are not as separate as once thought (Thapar & Rutter, 2016).

Prevalence

The target diagnostic disorders for this systematic review vary in prevalence. Williams Syndrome is the rarest with prevalence reported to be approximately 1 in 7 500 (Strømme, Bjørnstad, & Ramstad, 2002). Fragile X Syndrome is the most common genetic cause of inherited intellectual disability. Prevalence estimates for FXS are approximately 1 in 5 500 for males (Macpherson & Murray, 2016) and around 1 in 8 000 for females. However, prevalence estimates vary considerably especially due to advances in genetic testing (Hunter et al., 2014). The estimates for Down Syndrome prevalence have been reported in Europe and the US to be around 8 per 10 000 (Presson et al., 2013) whereas the prevalence for Autism Spectrum Disorder and Intellectual disability is considerably higher at approximately 1% prevalence in the overall population (APA, 2013).

Who delivers the intervention?

An important aspect of intervention research relates to who delivers the intervention. Evaluations of efficacy vs. effectiveness interventions where the former typically involves expert clinicians at for instance university clinics whereas effectiveness interventions mainly involve delivery of interventions in the child's preschool or school, delivered by the staff that work with the child on a day to day basis such as teachers, or by the parents of the child at home. Although efficacy trials are important it is also crucial for the possibility of broad implementation at community level that interventions can be delivered in ways that are manageable both in terms of cost and time efficiency. Thus, this review includes efficacy *and* effectiveness trials that are parent-implemented or delivered by persons working with the child at preschools/schools or in other more clinical settings.

To provide evidence for the effectiveness of language interventions for these children is important both in terms of what works in helping these children to reach their full potential, but also from a societal perspective to influence the development of policy and best practice for children with developmental disorders. It may be worth emphasising that interventions targeting language in children are plagued by lack of rigor, especially considering provisions of a sound theoretical rationale and evidence for efficacy (Hulme & Melby-Lervåg, 2015). Contributions to build a sounder evidence base in this field are therefore critical.

OBJECTIVES

The overarching aim of this review is to evaluate the effectiveness of interventions that set out to increase language skills in different groups of children with developmental disorders. This includes Autism Spectrum Disorders, Intellectual disability, Down syndrome, Fragile X, Language disorder, and Williams Syndrome. The main research questions are listed below:

How effective are language interventions for children across different developmental disorders?

Do the effects differ between groups of children with different disorders or problem profile?

Do the effects differ in terms of context of delivery of the intervention?

Are effects moderated by verbal and nonverbal intelligence?

What kind of interventions seems to be the most effective in enhancing language skills?

Are factors such as intervention duration, who delivers the intervention, and child age, related to intervention efficacy?

EXISTING REVIEWS

Below is a list of the reviews that is most closely related to the review we aim to do. However, as apparent from the list, there are no reviews that focus on broad inclusion of diagnostic groups in a cross-disordered manner. The lack of such pairwise comparisons is a gap in the literature since the focus has been on “pure” groups that to a lesser degree mirror real world clinical contexts (Bishop et al., 2016). Further, the need to investigate effects following interventions for these groups in relation to non-verbal IQ is highly warranted since explorations of the extent that such variables influences outcomes are scarce (Norbury et al., 2016). This is particularly relevant considering changes in diagnostic criteria and an inability to generalise previous intervention studies of children with language disorder to other clinical groups because studies of children with language disorders often exclude children with nonverbal IQ below 85. A note should be made about the ongoing Cochrane-review by Law and colleagues (2017*) that includes language disorder as is the case in the present title-proposal. However, although this is a slight overlap with the present proposal our review include children with “secondary” language difficulties, meaning that the problems in language are mainly caused by another condition or a disorder in contrast to the main focus of the Law et al. review that focus on speech and/or language disorders that have no known aetiology (i.e. “primary” difficulties in language (Law, Dennis, & Charlton, 2017)).

Cirrin, F. M., Schooling, T. L., Nelson, N. W., Diehl, S. F., Flynn, P. F., Staskowski, M., ... & Adamczyk, D. F. (2010). Evidence-based systematic review: Effects of different service delivery models on communication outcomes for elementary school-age children. *Language, Speech, and Hearing Services in Schools, 41*(3), 233-264.

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Hampton, L. H., & Kaiser, A. P. (2016). Intervention effects on spoken-language outcomes for children with autism: a systematic review and meta-analysis. *Journal of Intellectual Disability Research*, 60(5), 444-463.

Law, J., Garrett, Z., & Nye, C. (2005). Speech and Language Therapy Interventions for Children With Primary Speech and Language Delay or Disorder: A Systematic Review. *Campbell Systematic Reviews*, 1(5).

Reichow, B., Servili, C., Yasamy, M. T., Barbui, C., & Saxena, S. (2013). Non-specialist psychosocial interventions for children and adolescents with intellectual disability or lower-functioning autism spectrum disorders: a systematic review. *PLoS Med*, 10(12), e1001572.

Roberts, M. Y., & Kaiser, A. P. (2011). The effectiveness of parent-implemented language interventions: A meta-analysis. *American Journal of Speech-Language Pathology*, 20(3), 180-199.

*Law, Dennis, & Charlton (2017). *Cochrane Protocol: Speech and language therapy interventions for children with primary speech and/or language disorders*.

O'Toole, Lee, Gibbon, Bysterveldt, Paul Conway, Hart (2016). *Cochrane Protocol: Parent-mediated interventions to promote communication and language development in children with Down syndrome aged between birth and six years*. 2016

INTERVENTION

We aim to focus on language interventions that have been linked (theoretically and/or empirically) to positive language outcomes. This includes interventions that focus on enhancing skills like expressive or receptive language, listening comprehension, inference skills, or use of language in communication. The control condition should be a passive control group, active control group or waiting list control group. Studies with no control group will be excluded.

Note that there is a large set of literature that focuses on general cognitive training (such as working memory training, training of executive functions or auditory processing) for children with developmental disorders. The focus here is on interventions that target language skills and include language based tasks and outcome measures. Also, a number of interventions focus on language tasks that aim to improve children's phonological skills and/or articulation skills. The domains we will focus on here are studies that look at receptive and expressive vocabulary, grammar, listening comprehension, narrative skills, and we will exclude studies with interventions solely targeting the phonological domain.

We only include interventions that are directly delivered to the child, individually or in groups, from another person or persons. Methods of delivery will be in the preschools and schools (typically by a special education teacher, teacher, or assistants that have received targeted training related to the intervention) or by clinicians (typically by clinical staff such as Speech-language therapists). Also included are parent-mediated interventions. Interventions that involve only general teacher training are excluded from this review. Dietary interventions, pharmaceutical treatments, and other forms of non-person delivered

interventions (e.g. through computers and animal assisted interventions) are also excluded from the study. Further, interventions that target general cognitive skills are beyond the scope of this review.

POPULATION

We plan to consider interventions for children with developmental disorders that are known to display delays in both expressive and receptive language. This includes children with Autism Spectrum Disorder, Intellectual disability, Down syndrome, Fragile X, Language Disorder, and Williams syndrome. Studies of children having Specific Learning Disorder, impairments of speech related to oral-motor musculature such as articulation are excluded from the review as speech problems are distinct from language problems (Cohen, 2001). The age range will be 2-12 years comprising what would typically be the preschool and elementary/primary school years for typically developing children.

OUTCOMES

The primary outcomes are measures of the following constructs: expressive language (such as word definitions), receptive language (such as measured in Peabody Picture Vocabulary Test), listening comprehension (both grammar and discourse comprehension), mean length of utterance and narrative skills. We will also include studies where the outcome is pragmatic use of language in communication. However, note that for this outcome we will only include inferencing, figurative language use and discourse skills, i.e. measures that directly taps language skills. We will not include outcomes measuring more general social skills.

We will mainly focus on tests that assess language skills in children directly. We are including both standardised tests and custom made bespoke test materials. However, if direct tests are not available we will also include parental, clinician or teacher reports of language (such as MCDI). Assessment method can also potentially be an important moderator variable. As for other secondary outcomes when available, adaptive skills (e.g. Vineland Adaptive Behavioural scales (VABS)) will be of relevance. Further, measures of Nonverbal intelligence (NVIQ) will be used as an important moderator variable, as NVIQ poses as a potential critical factor for response to intervention for the target groups in this review.

STUDY DESIGNS

We aim to include quantitative studies that use a randomized experiment or a quasi-experimental design with a control group. The studies have to include baseline measures. Quasi-experimental designs with control groups are included in the review as it would otherwise be difficult to build a large enough pool of studies from which to build recommendations on.

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ROLES AND RESPONSIBILITIES

- **Content:**

All authors have experience in topics related to language and developmental disorders and intervention

- **Systematic review methods:**

Professors Norbury, Melby-Lervåg, and Lervåg have published several systematic reviews in international peer reviewed journals and have expertise within systematic review methods

- **Statistical analysis:**

Professors Norbury, Melby-Lervåg, and Lervåg have published systematic reviews deploying meta-analytic statistical analyses in international peer reviewed journals.

- **Information retrieval:**

The review team has experience with electronic database retrieval. The authors will collaborate with Information retrieval expertise at the library of University of Oslo when needed.

FUNDING

This project receives financial support from the Research Council of Norway (Educational research 2020)

POTENTIAL CONFLICTS OF INTEREST

None declared

PRELIMINARY TIMEFRAME

- Date you plan to submit a draft protocol: 30 February 2017
- Date you plan to submit a draft review: 15 August 2017

AUTHOR DECLARATION

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