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Defeating dyslexia: A robust meta-analysis of the relationship between hearing and dyslexia to achieve early diagnosis

By Rick Qian[1]

[1]*Northwestern University*

Abstract: Occurring in nearly 20% of children, dyslexia is all too often misdiagnosed or overlooked. Untreated, it can irreversibly disrupt the early learning period, preventing students from reaching their full potential. Thus, early and effective intervention is critical. Falling behind as a dyslexic creates challenges not only academically, but also emotionally and financially. The purpose of this study is to quantify the relationship between dyslexia and deficits in sound gap detection, expanding on the viability for hearing tests to support early diagnosis.

The relationship between dyslexia and auditory processing in gap detection is examined using the tool of meta-analysis. Data collected from 15 studies was analyzed with REDCap and R, and a Hedges' *g* test and Robust Variance Analysis was performed on the data set. Poor ability to detect gaps in sound were significantly correlated to dyslexia through both well-established and novel methods. This study is the first work to specifically focus on gap detection in dyslexics, and the strength of relationship ($g = 1.17$) demonstrated in this study is one of the highest reported among meta-analyses for auditory processing tasks.

These findings could influence how future diagnosis and treatment of dyslexia advances. Early detection is key to effective treatment, and auditory testing is viable at pre-reading ages. Auditory processing as a major deficit in reading disorders also supports the hypothesis that hearing is the fundamental cause of dyslexia, affecting future treatments to address auditory issues more directly and bringing the field one step closer to defeating dyslexia.

Introduction

Dyslexia is the most common learning disorder in the world, occurring in 1 out of 5 children and representing 90 percent of all learning disorders in the US (Shaywitz, 2017). Without early and effective treatment, dyslexia can have serious long-term consequences. Students with undiagnosed dyslexia fall behind in school, unable to develop key reading skills at an early age. These frustrations have devastating psychological impacts, and untreated dyslexics are at much higher risk for depression, anxiety, and poor self-image (Richardson & Wydell, 2003). Higher education becomes an increasing challenge (Humphrey & Mullins, 2002), closing off dyslexics from hundreds of careers and thousands of dollars in future income.

Dyslexia is characterized by difficulties with accurate and fluent word recognition. Signs like delayed speech and inability to recognize rhyming words are strong indicators of dyslexia and can be apparent at an early age. Most dyslexia deficits are due to difficulties in phonological processing, which impede the ability of a child with otherwise normal intelligence to speak, read and spell. Secondary deficits may include problems in reading comprehension and reduced reading experience that can impede growth of vocabulary and background knowledge.

Research on the causes of dyslexia focuses on two major categories of deficits, phonological and auditory perceptual. Phonological processing is impaired in dyslexics in their ability to recognize and break down words into syllables and decompose syllables into small distinguishing sounds called phonemes. Phonological deficit theory is a top-down methodology; it theorizes difficulties in phonological processing such as in syllable and phoneme recognition are a result of higher-level deficits for phrase and entire word processing (White et al., 2006). These same impairments translate to difficulties in Rapid Automatized Naming, which result from slow recognition, storage, and retrieval of given phrases.

Two main strategies are typically used to develop the phonological skills necessary for reading: the logographic strategy and the alphabetic decoding strategy (The Open University). The logographic strategy involves the association of entire words with their written forms, and it places huge demands on visual memory. To address these limitations, children also utilize an alphabetic decoding strategy, where they learn the sound each letter of the alphabet makes, and then learn to blend them together during reading to work out the pronunciation. Both strategies are impaired in dyslexics, as shown with their difficulty in decoding, storing, and recombining words and syllables. Phonological deficits play a major role in causing dyslexia and the conditions that make literacy so challenging (Snowling, 1995). These specific deficits make phonics one of the most effective ways for dyslexics to learn literacy skills, as it specifically focuses on teaching dyslexics to blend individual syllables and letters, allowing for entire words to be recognized and decoded. Phonics serves as the primary treatment method for dyslexics struggling to read.

The area of research concerning auditory processing in dyslexics is an alternative theory to the phonological viewpoint. Significant findings have been noted between deficits in auditory perception and dyslexia, such as in frequency discrimination (Witton et al., 2020) and duration discrimination (Leong 2013). These connections between auditory processing and dyslexia have led theories on auditory deficits to play a more fundamental role in dyslexia. The auditory processing perspective is a bottom-up approach, where basic auditory deficits are seen as the cause of phonological deficits. One study suggests that speech and sound processing lead to inaccurate recording of words, greatly diminishing phonological ability (Pasquini et al., 2007). Phonological deficits are shown to be correlated with deficits in auditory processing (Thomson 2009), giving credence to such possible theories.

A promising avenue in understanding the cause of dyslexia is through a neurobiological lens. One theory proposes that the disruption of auditory perception and difficulties in processing linguistic input occurs at the cortical level, above periphery systems of hearing on the outer layer of the cerebrum. Advancements in neuroscience and MRI technology will allow greater study of neurological areas of interest, both in auditory theories and phonological based theories, and several studies (Norton, 2016; Elnakib et al., 2014) have already begun exploring this area. There are many theories on the true nature of auditory processing deficits in dyslexics and much uncertainty regarding dyslexia's role in those deficits, which this study seeks to reduce.

Objectives

The purpose of this meta-analysis is to assess the association between auditory processing deficits and dyslexia (RD). Specifically, I examine the relationship between auditory temporal resolution, as measured by gap detection, and dyslexia. Included studies must possess several characteristics: an original study that is not a case study (n>5), a group of children with RD, a control group of typically developing children (both groups must have begun formal schooling), a relevant behavioral auditory processing task, a calculable standardized mean difference, pure, noise, or complex tones (no speech or syllable stimuli), and two tones, or sounds, per trial. Data from relevant studies will be extracted and stored in a centralized database.

Hypothesis

The hypothesis investigated in this study is that reading impairments due to dyslexia are correlated with auditory temporal processing deficits. A meta-analysis will be conducted to determine the statistical relationship between dyslexics and their ability to detect gaps in auditory stimuli.

Materials and Methods

Identification of Studies and Data Collection

61 relevant articles were identified with Google Scholar, Medline, and PsycINFO using a systematic review conducted by Hamalainen et. al 2013. As part of a larger meta-analysis, 4770 potentially relevant sources were identified through snowball searching. Snowball searching is essential to identify all relevant effect sizes in the literature that should be included in analysis.

Of these 4770, 2215 were screened in abstrackr software, and the 845 articles that qualified were full text assessed for eligibility. In order to qualify for full-text screening, coders must answer in the affirmative to each question below:

- Is the study an original research study (i.e. not a meta-analysis, review, or editor's commentary)?

- Does the study have several participants (i.e., not a case study; n>5)?

- Are the participants old enough to be diagnosed with dyslexia? (mean age ≥ 6; begun formal schooling)

- Does the sample comprise typically developing individuals other than reading and language difficulties (e.g., not deaf or blind, no ASD, but could include ADHD)?

- Does the abstract mention frequency discrimination, intensity, duration, or gap detection?

- Does the abstract mention learning disability, reading disability, or dyslexia?

Of these 845 articles, 15 met the full eligibility criteria for study inclusion and had reported means and standard deviations from which to calculate an effect size and sample variance.

Study Inclusion

Study inclusion methods examined each of the 15 studies with the following requirements: No study was excluded because its effect size is an outlier unless the study should be excluded for poor study quality. Models with extreme outliers (≥2 SD) both included and excluded were presented if such outliers are present. Studies that use alternate terms such as dyslexia or "poor readers" were included, given the heterogeneity of definitions used in both research and practice that use different types of reading measures and scores for inclusion and for a formal diagnosis. Studies were excluded if the study sample is designed for autism spectrum disorder, schizophrenia, or chromosomal disorder. If the study does not list their participant exclusion list, it may have individuals with autism spectrum disorder (ASD) incidentally and was included.

Studies examined required that children had begun formal reading instruction and thus would be able to be diagnosed with reading disorder (RD). This is primarily because approximately 50% of children who are identified as at-risk for reading problems based on family history of reading problems will not go on to develop RD, thus diluting the analysis. Studies were excluded if they do not measure auditory processing and/or the primary auditory domain I am interested in. Studies were also excluded if they were written in a language other than English, a duplicate of a previous record, or if the full text file could not be located after extensive searching.

Data was collected on the demographics of the samples, study measures, effect sizes for gap detection, and study quality measures. All data collection was done using the Northwestern REDCap system, with manual coding to enter in the data identified. The age, primary language, sample size, exclusion of children with attention deficit hyperactivity disorder (ADHD) and/or developmental language disorder (DLD), and bilingual status was extracted.

Study measures included information on the specific tasks performed to test gap detection, such as whether adaptive or fixed methods were used, the task design, frequency, wavelength, intensity, or gap length of the auditory stimulus. The possible task designs are described below:

Two-alternative forced-choice (2AFC): Requires participants to choose between two stimuli on the domain of interest (e.g., which tone was higher/lower, longer/shorter, or louder/quieter).

Same-different tasks: Uses two stimuli but only requires the participant to respond whether the stimuli were the same or different (similar to the yes-no method).

Three-alternative forced-choice (3AFC): Requires participants to select the “odd one out” from a group of three sounds.

AXB task: Also uses three sounds, but the middle sound is a fixed reference, and the listener chooses between the first and last sound on the domain of interest.

ABABA/AAAAA task: Uses 10 stimuli over two intervals and participants are asked which interval had two different sounds.

Two interval, two-alternative forced-choice (2I-2AFC): Uses four sounds in two intervals and participants will be asked which interval (1 or 2) had the differing sounds.

Gaps-In-Noise (GIN) test: Uses a series of uniformly distributed noise segments, each of which contains zero to three silent gaps varying in duration, and the participant is tasked with identifying the gaps in sound.

Fusion Task: A series of auditory stimuli will be played in sets of one or two, and the participant will be asked if they heard two sounds or one.

Effect sizes were collected on the “just noticeable difference” (JND) threshold data reported for the gap detection task, defined as the minimum silent gap between two stimuli that a person can detect reliably. “Reliably” ranged from answering correctly 70.7 percent of the time (Levitt 1971) to 79.4 percent of the time depending on the study. The mean and standard deviations of the threshold were recorded for this. Additionally, a Cohen’s d test was performed using the Becker online calculator and logged into REDCap as a verification for the Cohen’s d test performed later in the statistical analysis.

Study Quality

Study quality was evaluated with a 14-question survey to determine the quality of its data. The questions were:

(1) Was the research question or objective in this paper clearly stated?

(2) Was dyslexia/reading disability clearly defined using cutoff scores and appropriate measures? Was the control group free of RD subjects?

(3) Was the participation rate of eligible persons at least 50%?

(4) Does the dyslexia group have the same inclusion/exclusion criteria other than reading performance?

(5) Was a sample size justification, power description, or variance and effect estimate provided?

(6) Were dyslexics recruited from the same source as typical participants?

(7) Was the timeframe sufficient so that one could reasonably expect to see an association between exposure and outcome if it existed?

(8) Did the study measure reading performance for typical and dyslexic subjects alike?

(9) Were the tests used to measure dyslexia (and other subgroupings, such as ADHD) defined in detail?

(10) Were the participants' reading skills (or dyslexia status) assessed more than once over time?

(11) Were the auditory processing tasks clearly defined, valid, reliable, and implemented consistently across all study participants?

(12) Were the assessors blind to the participants' diagnosis?

(13) Was loss to follow-up after baseline 20% or less?

(14) Were key potential confounding variables (e.g., attention or language impairments, age) measured and adjusted statistically for their impact on the relationship dyslexia and auditory processing?

The answers to this survey guided the final decision to mark the study as either poor, fair, or good quality. Assessment of the quality of selected studies is a useful metric for qualitatively weighing the statistical value of each study.

In addition, an Egger's Sandwich Regression (Egger et. al 1997) test was performed to assess the publication bias of each study, or the bias that results when conclusions affect the probability of the study being published. Egger's test is a funnel plot for the linear regression of the effect of inverse variance compared with treatment effect size. The funnel plot used in this study will have the treatment effect size and study precision on the x and y axis, respectively. A funnel plot depicts a "well-behaved" data set and funnel plot asymmetry indicates an association between treatment effect and study precision, suggesting the possibility of publication bias. The Eggers' Regression tests used data from independent samples only; that is, data from the same study will be aggregated to find the overall bias value. This is done to remove outliers not representative of the entire study, preventing the funnel plot from being unreasonably skewed.

Data Analysis

Data analysis for this meta study was done using Excel spreadsheets and the coding language R. R was used to pull effect size data from the REDCap system on each of the identified relevant studies into arrays and aggregate all dependent data. Gap detection tasks from the same study, such as left ear and right ear assessment, were considered to be highly correlated and dependent data, which would be handled by our meta-analysis models described below. Effect size was calculated from each study's group means, SDs, and sample sizes using Hedges' g values. This was done with the escalc function from the metafor R script package.

Hedges' g is a standardized mean difference effect size measure that corrects for small sample sizes ($n<20$). Variance for these g values were also estimated using the R package metafor and quantified with 95% confidence intervals. Low variance indicates higher precision which will more heavily weight its associated g.

A Robust Variance Estimation (RVE) model using the metafor package in R was performed using the Hedges' g values and their corresponding sample variances, accounting for the statistical weight of each study. This gave the overall effect size, confidence interval, heterogeneity values, and t test results at $p = 0.001$. RVE models allow for correlated tasks (i.e., tasks performed by the same participants) to be analyzed without violating assumptions of independence. This allows for all possible effect sizes to be run in a single meta-analysis model. The method can also be applied in situations where correlations arise because of study-level characteristics (such as shared investigators or laboratories), sometimes called the hierarchical dependence model.

Results and Discussion

Hedges' g test

Hedges' g effect size values for each study varied across a range of values from 0.179 to 5.31, though most values were below 3.34. Hedges' g 95% confidence intervals were roughly consistent, with no study having a particularly small or large range. The difference between Hedges' g and Cohen's d was also calculated, with Cohen's d values resting within 0.272 above or below the Hedge's g value. A Hedges' g test was done for each dependent variable and the data table below shows the final effect size after aggregating multiple effect sizes if present in a study into a single effect size. A 95% confidence interval was generated around the effect sizes and interval size increased when studies had high variance and SD values. A Forest plot illustrated the confidence intervals for each respective study (Supplementary Materials, Table 1).

The effect sizes and age of the sample (adults vs. children) had no discernible relation between them, though adults and adolescents typically had much higher base threshold values when looking at the sample means. Larger sample sizes typically had much smaller standard deviations and variances while effect sizes changed irrespective of the sample size.

Gap Detection Measures (Robust Variance Analysis)

A Robust Variance Analysis using the Hedges' g effect sizes on the entire data set was performed, giving a mean effect size of 1.17, and a t value of 3.77 at p = 0.001. The 95% confidence interval was 0.51 - 1.82. There was a large amount of heterogeneity (I^2 = 90.85 (bound between 0-100), Tau^2 = 1.16) in the data set, showing the high diversity of data values in the studies examined.

Eggers' Regression Test

The Eggers' Regression test for Random effects gave a p value of 0.07, which was non-significant. An Eggers' Sandwich Regression test where the values were not aggregated based on individual studies gave a p value of 0.11, also non-significant.

The standardized mean difference of each study was correlated with their standard error on the funnel plot, and values are within or near the funnel. The funnel plot was created around the mean of the samples taken, and all values are aggregated, independent samples (Supplementary Materials, Table 3).

Limitations

This study and meta-analyses in general have several types of possible errors. These include publication bias in the sample studies, low quality of data reported in the studies, confounding variables in the studies themselves, and small sample sizes. These were addressed with an Eggers' Regression test, study quality assessment, and heterogeneity calculations in the data. Publication bias was shown to be non-significant with the Eggers' Regression test and the quality of reported studies was considered overall to be medium as per the study quality survey. This shows that the data in the studies selected was reported with minimal bias for positive results and the accuracy of that data was moderate, as most studies had clearly defined thresholds and measures for auditory processing and dyslexia. However, no study adjusted for confounding variables or justified their sample size, lowering the accuracy of results. A Hedges' g test was selected for this study over a Cohen's d test to fit the small sample sizes found in selected studies (n<20). High heterogeneity for individual studies indicates the presence of confounding variables, such as nonstandardized tests, diverse task designs, and differences in the definition of dyslexia and ages of study subjects, leading to less precise measurements of effect size and greater variance. This is consistent with the absence of adjustment for confounding variables in any study. These issues can be corrected in future studies with more stringent study inclusion criteria and assessment of study quality after a publication has been full text coded, which increase the chance of selecting high quality studies.

Conclusions

This study's primary focus is investigating the link between dyslexia and auditory processing deficits in gap detection. The average gap detection deficit in individuals with dyslexia was large ($g = 1.17$; 95% CI: 0.51-1.82). Effect sizes estimates above 0.8 are considered large (Lakens, 2013), and our results greatly exceed that benchmark.

Another more optimal approach discussed by that study is to relate the effect sizes of the target study to those of other studies in the literature and explain the implications of those effects. The large effect size in this study is supported with another meta-analysis (Araujo & Faisca, 2019), which describes the effect size between Naming Speed Deficits and dyslexia.

Naming speed measures how fast a subject can retrieve a symbol and is theorized to form another core deficit of dyslexia, which creates the fluency issues that often result in dyslexics.

The similar effect size in Araujo & Faisca, 2019 ($d=1.19$) may indicate a shared underlying neural mechanism, with auditory and fluency deficits as outward signs of a root problem. One approach addressing these similarities is the temporal sampling framework (Goswami, 2010). The framework proposes a model of dyslexia from a syllabic, rather than a phonemic perspective when looking at phonological development. Syllabic perception, described as the ability to recognize the syllabic structure of words, is often impaired in dyslexics.

Impairments in this area are often coupled with difficulty recognizing the prosodic, or sound structure of each syllable. This syllable level representation has elements of both phonological and auditory deficits, fitting well with the relation seen between dyslexia and deficits in both processing gaps in tonal sound and processing the syllabic structure of words (Araujo & Faisca, 2019).

The temporal sampling framework contrasts with the traditional phonological viewpoint of dyslexia as a deficit in detecting phonemes in phrases, theoretical sounds that form the difference between close sounding words, such as Cat and Hat (Goswami, 2010). Children develop phonemic awareness at pre-reading ages, learning to isolate and blend sounds into words to learn to read and spell. Dyslexics are unable to do so during reading development and require explicit instruction to begin acquiring such skills. This is a segment level approach to the disorder that could be too specific when considering the variety of deficits around dyslexics and does not explain the correlation between the two studies as well.

These auditory and phonological deficits may be fundamentally rooted in an impairment of the "phase-locking" ability when listening to modulated noise. Phase locking is the tendency of neurons to fire at certain frequencies when exposed to an auditory stimulus as a way for the brain to perceive sound (APA Dictionary of Psychology). One study investigated the effect of modulating frequency, amplitude, and time length on phase locking, and results showed a significant change in each category (Gransier et al., 2021). For non-dyslexics, differences in the time length of gaps between noise are corresponded with changes in frequency of neuron action potentials; these same changes may be much less pronounced in dyslexics, leading to their poorer ability to detect gaps in auditory stimuli.

Future Work

Overall, the analysis performed in this meta study displays a clear relationship between dyslexia and deficits in auditory gap detection, though whether this relationship is causal, or the result of confounding factors requires further investigation. My next steps are exploring a broader scope of auditory processing tasks, including frequency and intensity discrimination deficits, bringing a more complete picture of auditory processing deficits and dyslexia. An area for future study is the underlying deficits behind auditory processing, such as phase locking, to establish the possibility of a causal relationship. This could enhance understanding of dyslexia and steer treatment methods to address the auditory component of the disorder. The continual improvement of diagnosis, treatment, and our understanding of dyslexia through first-hand studies and meta-analyses brightens the prospects of millions of dyslexics across the world. With these efforts in place, the foundations of ongoing and future dyslexia research are strengthened, taking one step forward in the search to defeat dyslexia.

Supplementary Information

Study	Effect Size	RD threshold value/SD	RD/ Control sample size	Control threshold value/SD	95% Confidence Interval	Age Category
Boets et al., 2011	0.56	0.522/0.267	16/46	0.422/0.132	-0.0156-1.14	Child
Breier et al., 2003	0.053	2.8/2	76/41	2.7/1.6	-0.327-0.433	Child
Chaubet et al., 2014 **(Left Ear)**	1.3	7.3/2.23	26/21	5/0.89	0.650-1.91	NA
Chaubet et al., 2014 **(Right Ear)**	1.27	7.5/2.97	26/21	4.6/0.57	0.640-1.90	NA
Fostick et al., 2012	5.3	15.33/1.06	37/40	9.76/1.02	4.36-6.25	Adult
Gokula et al., 2019 **(Left Ear)**	0.83	6.36/1.93	25/28	5.14/0.8	0.269-1.39	Child
Gokula et al., 2019 **(Right Ear)**	0.94	6.36/1.93	25/28	4.96/0.88	0.370-1.51	Child
Ingelghem et al., 2001/2005	1.39	3.3/0.5	10/10	2.7/0.3	0.416-2.37	Child
King et al., 2003	0.60	15/9.3	11/14	10.8/3.8	-0.207-1.41	Adult
Mccroskey & Kidder, 1980 **(7-year-olds)**	0.76	14.7/9.25	15/15	8.9/4.85	0.0231-1.51	Child
Mccroskey & Kidder, 1980 **(8-year-olds)**	0.38	9.9/4.93	15/15	8/4.93	-0.347-1.01	Child
Mccroskey & Kidder, 1980 **(9-year-olds)**	0.60	11.7/9.18	15/15	7.5/3.09	-0.135-1.33	Child
Schulte-Korne et al., 1998 **(Adults)**	0.18	3.2/0.7	9/22	3/1.2	-0.597-0.956	Adult
Schulte-Korne et al., 1998 **(Children)**	0.34	11.3/5	15/14	9.6/4.8	-0.397-1.07	Adolescent
Schulte-Korne et al., 1999	0.55	14.1/10.3	19/15	9.4/4.7	-0.138-1.24	Adolescent
Sharma et al., 2006 **(Click)**	0.93	15.7/8	15/19	9.4/5.3	0.217-1.64	Child
Sharma et al., 2006 **(Tone Average)**	0.84	12.6/8.3	15/19	7.5/3	0.133-1.55	Child
Vandewalle et al., 2012	0.90	110.1/86.9	8/14	58.4/25.9	-0.0119-1.80	Child
Zaidan & Baran, 2013 **(Left Ear)**	3.3	8/1.46	31/30	4.27/0.52	2.56-4.11	Child
Zaidan & Baran, 2013 **(Right Ear)**	3.2	8.48/1.73	31/30	4.2/0.61	2.47-4.0	Child
Zaidan, 2009 **(Right Ear)**	2.2	6.7/1.3	61/30	4.2/0.6	1.67-2.75	Child
Zaidan, 2009 **(Left Ear)**	2.3	6.5/1.1	61/30	4.3/0.5	1.75-2.85	Child

Table 1. Effect size, means, and standard deviations for reading disabled (dyslexics) and controls.

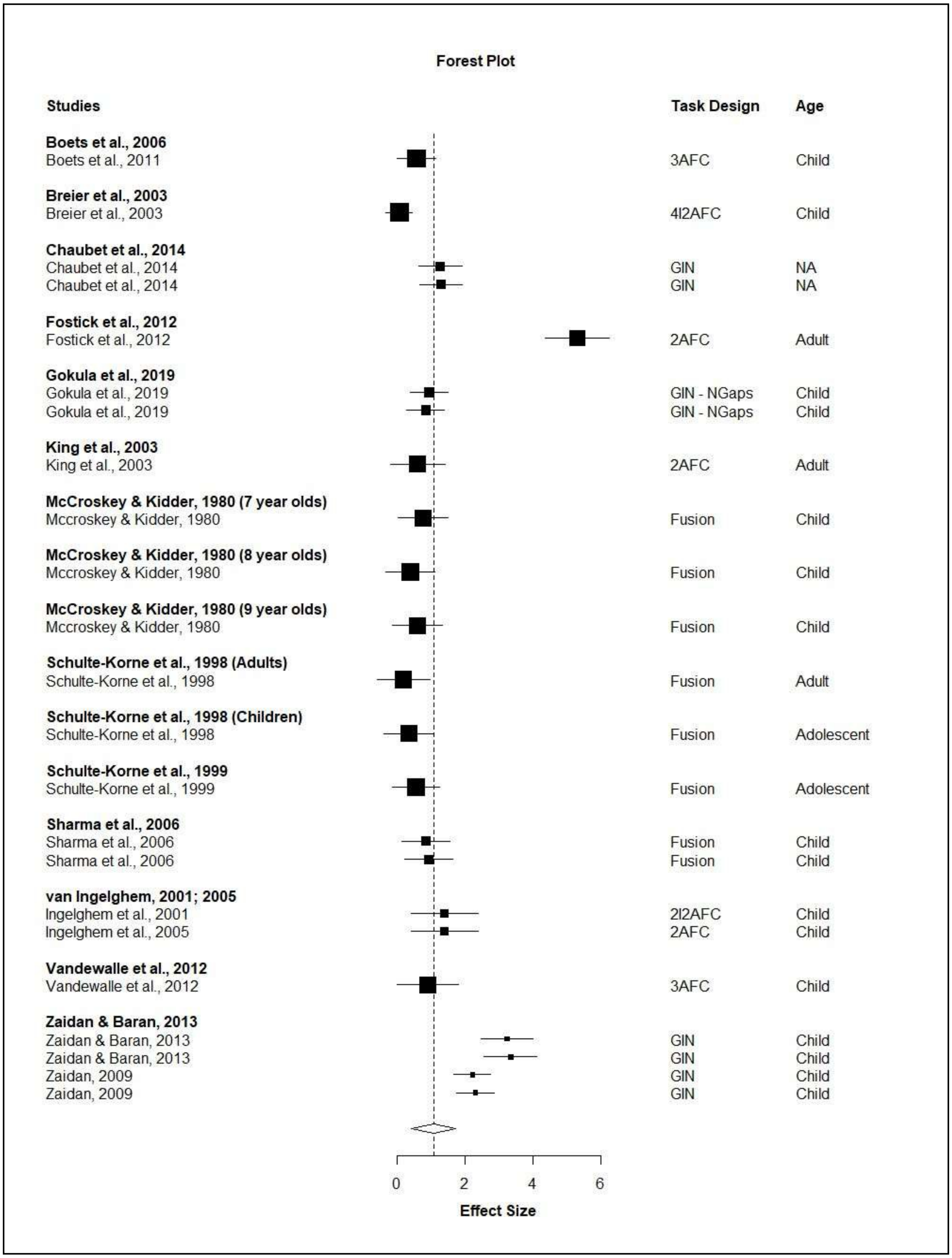

Table 2. Forest plot for confidence intervals in each study.

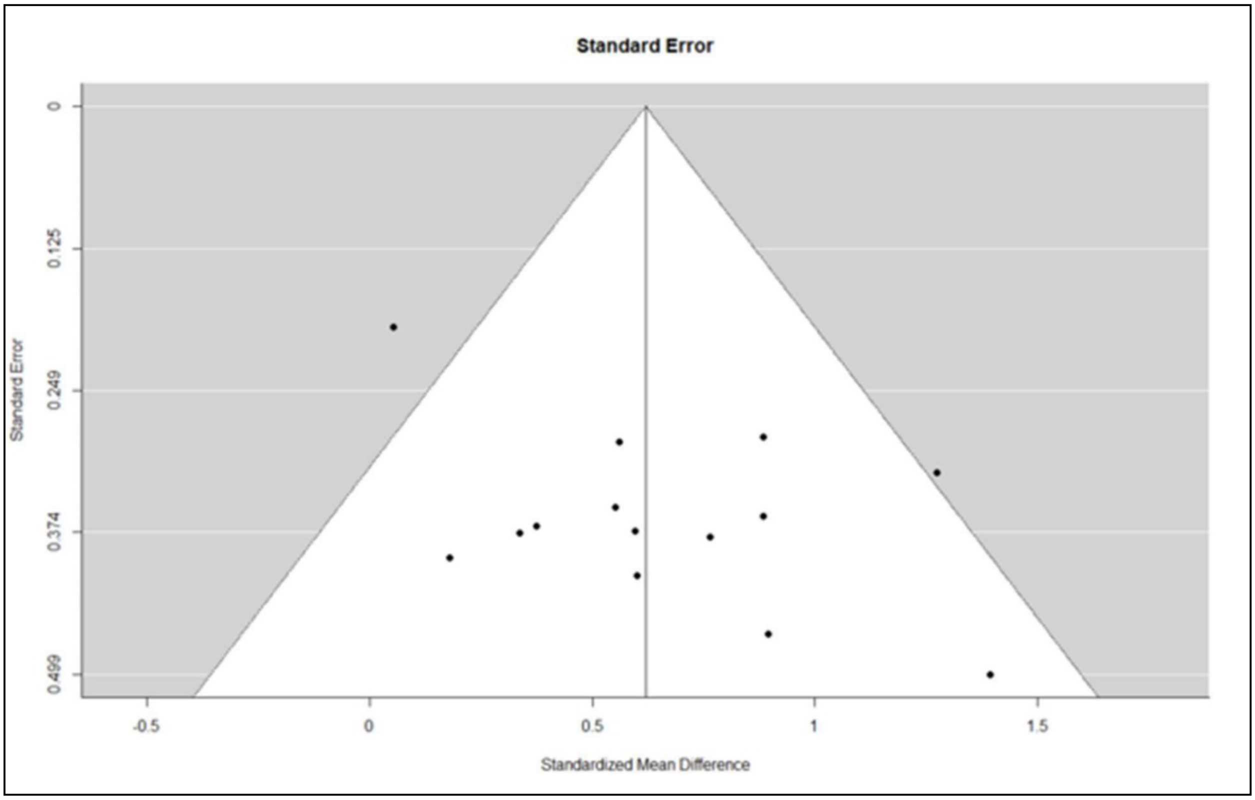

Table 3. The standardized mean difference of each study was correlated with their standard error on the funnel plot, and values are within or near the funnel. The funnel plot was created around the mean of the samples taken, and all values are aggregated, independent samples.

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