Motor-based interventions improve language outcomes in children with autism: a systematic review

Christina E. Odeh¹, Rebecca Martell², Sarah Griffin¹, Erik R. Johnson³, Allison L. Gladfelter³

¹Physical Therapy, School of Allied Health & Communicative Disorders, Northern Illinois University, DeKalb, IL; ²School of Interdisciplinary Health Professions, Northern Illinois University, DeKalb, IL; ³Speech-Language Pathology, School of Allied Health & Communicative Disorders, Northern Illinois University, DeKalb, IL, USA

Purpose: Children with autism spectrum disorder (ASD) show motor deficits in addition to the social communication and repetitive behaviors characteristic of the disorder. However, these deficits have traditionally been targeted independently during intervention. The primary purpose of this systematic review was to determine whether interventions with motor targets improved language or social communication outcomes in individuals with ASD.

Methods: Five databases were searched using the following terms: autis*, asper*, motor*, therap*, interven*, and treat*. After eliminating irrelevant and duplicate articles, 74 articles underwent full text review to determine whether they met the inclusionary/exclusionary criteria. The 15 included articles were then checked for inter-rater reliability and appraised for the quality of their research design, treatment fidelity, and interobserver agreement. Following the quality appraisals, 13 included articles were analyzed for final data extraction.

Results: Of the 13 included studies, 12 showed at least one increased language outcome, seven demonstrated at least one increased motor outcome, and one revealed no significant change in either language or motor outcomes.

Conclusions: Consistent with previous research, many of the children with ASD presented weaknesses in both motor and language skills. In most studies, the motor-based interventions led to an increase in language skills, indicating language and motor system interdependence. These findings also suggest that co-treatment between physical therapists and occupational therapists alongside speech-language pathologists may be warranted when working with children with ASD.

Keywords: Autism, Motor intervention, Language outcomes

INTRODUCTION

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterized by persisting impairments in social communication and interaction as well as restricted and repetitive behavior, interest, and activity patterns presenting in early development [1]. There is currently no motor-related diagnostic criteria other than repetitive, stereotypic movement patterns, such as rocking or hand flapping [1]. Motor abilities implicated in children with ASD vary significantly. The presence of motor involvement in children with ASD is widespread, with up to 79% of children with ASD presenting with
motor performance deficits [2]. Although motor impairments are known to present alongside communication weaknesses as a pervasive feature among children with ASD [3], motor and communication deficits traditionally are targeted independently of one-another. A shift from tradition toward a more integrative treatment approach may be warranted, as a growing body evidence suggests not only a relationship, but a level of interdependence between motor and language systems is present [4,5].

A critical relationship between the developing motor and language systems in children with ASD has been made evident. The evidence of relationship between motor and language development is demonstrated in the multiple findings which illustrate that motor function during infancy and toddlerhood is predictive of later communicative skills in children with ASD [6-8]. Stone and Yoder [8] found that motor imitation skills during toddlerhood is predictive of later preschool-aged speech fluency in children with ASD. Poon and colleagues [7] observed that a delay in the development of imitation and object play skills is predictive of communication ability by age 3. Furthermore, motor skill at 6 months of age was found to predict expressive language at 30 and 36 months of age in children with ASD [7]. LeBarton and Landa [6] even suggested that early inclusion of motor intervention may serve to mitigate the impact of motor delay on later social and communicative skills in children with ASD.

The intricacy of connection between developing motor and language systems is not fully understood; however, recent evidence has revealed that differences in ability between domains of motor learning may provide differing contribution and impact to communication and language development in children with ASD. Mody et al. [9] suggested a difference in influence of gross versus fine motor abilities on communication profiles in children with ASD. In their retrospective study, for children with ASD aged 2-17 years, it was found that fine motor scores on the Mullen Scales of Early Learning (MSEL) increased, MSEL receptive and expressive language scores also increased. Similar findings were not present when gross motor abilities were examined; a lack of relationship between gross motor and expressive language as well as a negative relationship between gross motor and receptive language was found. The authors went on to speculate that impairment in gross motor functioning may affect the development of joint attention abilities, possibly accounting for a resultant difficulty attending to speech impacting receptive language development.

Despite the need for a more complete understanding of the breadth and intricacy of the relationship between motor and language development in children with ASD, it is evident that a complex relationship exists. The presence of interactivity between motor and language systems necessitates consideration for both systems in approach to diagnosis and treatment. There is an emerging body of evidence indicating a relationship and degree of interactivity between developing motor and language systems in both neurotypical children and children with ASD [10]. This implicates the need for further investigation to determine whether incorporation of intervention targeting motor function influences outcomes in language and social functioning for individuals with ASD. The primary objective of this systematic review was to determine whether interventions targeting motor function facilitate improvement in language outcomes in individuals with ASD.

METHODS

Inclusion criteria

In order to be included in this review, the participants had to meet the following criteria: 1) presented with Autism Spectrum Disorder, PDD-NOS, or Asperger’s Syndrome according to the DSM-IV or V; the International Classification of Diseases, the Autism Diagnostic Observation Schedule or the Autism Diagnostic Interview-Revised, 2) had not begun or altered a pharmacological intervention within three months of the onset of the experiment, 3) did not present a concomitant diagnosis of a genetic, cognitive, or hearing impairment (e.g., Fragile-X, Angleman, etc.) at the start of treatment, and 4) were not labeled as bilingual learners of language. The studies reviewed had to also meet the following criteria: 1) reported outcomes for at least one language-based measurement, 2) included a treatment or intervention; the study may have evaluated one treatment or compared two or more treatments, 3) focused on a motor-based treatment, 4) did not use graphic symbols as a sole means of communication (such as early phases of Picture Exchange Communication System), 5) employed a (quasi-) experimental design for evaluating the effectiveness or efficiency of a treatment, such as selected (quasi-) experimental group designs or single-subject experimental designs (pre-experimental designs such as AB-designs or group equivalents were excluded), 6) was written as an article in a refereed journal, a book chapter, or a document made available through the ERIC; or it appeared in published conference proceedings or as an unpublished Master’s thesis or doctoral dissertation, and 7) the experiment was dated be-
between 1989 and December 2019 (including those that are published on-line first).

Search strategy
The following electronic databases were searched: CINAHL, Web of Science, PsycINFO, Cochrane. The following key words were used when searching for articles: autis*, asper*, motor*, therap*, interven*, and treat*. Using these search terms, the initial title hits resulted in 5,584 articles. Then, abstracts were assessed to further compare appropriateness of inclusionary/exclusionary criteria. Following the abstract review and after eliminating duplicate articles, 74 articles underwent full text review to determine whether they met the inclusionary/exclusionary criteria.

Full-text review was completed by an undergraduate student majoring in pre-physical therapy and a speech-language pathology master’s student. All 74 articles underwent an inclusion/exclusion criteria process, in which the study had to meet all 11 check points on the inclusion checklist to be included in the systematic review. Following the full-text review, 15 articles remained in the systematic review.

Interrater reliability for inclusion criteria
Coding on 37 (roughly 50%) of the 74 articles was also applied, blindly, by a speech-language pathologist with a PhD to determine the interrater reliability of the application of the inclusion/exclusion criteria process. Interrater agreement before consensus building was 91%, Kappa = 0.83, Phi = 0.84. Disagreements were resolved through a consensus process.

Guidelines for study evaluation
A study quality appraisal rating was completed on each study following the guidelines put forth by Simeonsson and Bailey [11]. This system classifies evidence into four categories: **conclusive**, **preponderant**, **suggestive**, and **inconclusive**. These categories are based on three types of information: research design, interrater reliability (IRR), and treatment fidelity (TF). A **conclusive** classification includes articles that have a clear, sound research design, adequate IRR and TF, and provide outcomes that undoubtedly resulted from the intervention implemented. A **preponderant** study showed minor flaws in either the research design or in the IRR or TF reported and presented outcomes that are likely to have occurred due to the intervention. **Suggestive** studies reported flawed or missing IRR or TF, some minor design flaws, and resulted in outcomes that could possibly be drawn from the intervention. Finally, an **inconclusive** study had either fatal design flaws or had missing IRR and TF and showed that no conclusion could be made about the outcomes of the intervention. The study quality appraisal ratings were completed by an undergraduate student majoring in pre-physical therapy. The quality appraisal rating for each included study is presented in Table 1.

Interrater reliability for quality appraisal ratings
Five (33%) of the 15 included articles were then checked for inter-rater reliability of the appraised quality of their research design, treatment fidelity, and interobserver agreement by a blind, second coder with a PhD in speech-language pathology. The interrater agreement was 100%.

Data extraction
Following the quality appraisals, 13 included articles were analyzed for final data extraction using an agreed upon coding manual. The two articles (of the original 15 included) that did not undergo coding were classified as inconclusive. The study characteristics coded included: author, year of study, participant age, participant gender, participant diagnosis, measures used to assess baseline, intervention design, treatment used, intervention provider, intervention outcomes, interobserver agreement, treatment fidelity, and effect size.

Interrater reliability for data coding
The first round of data coding was completed by an undergraduate student majoring in pre-physical therapy. For IRR, 4 articles (30% of total) were randomly selected to be coded by a graduate student studying physical therapy. The second coding was done blindly. The resulting IRR was greater than 80% for all four articles, with a range of 83% to 95%, resulting in an average IRR of 87%. Disagreements were resolved through a consensus process.

RESULTS

Participant characteristics
At least part of the participants in an included study must present a diagnosis of Autism Spectrum Disorder according to the DSM-IV, DSM-V, ADOS, or ADI-R. A total of 253 children between the ages of 22 months and 16 years of age participated in the studies included.

Research design
Five studies implemented lower quality randomized con-
Table 1. Summary of studies that examined language outcomes with motor interventions

<table>
<thead>
<tr>
<th>Authors</th>
<th>Participants</th>
<th>Goal of intervention</th>
<th>Type of intervention received</th>
<th>Language outcomes</th>
<th>Motor outcomes</th>
<th>Appraisal</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ingersoll (2012)</td>
<td>M: 24, F: 3</td>
<td>Increase participants’ language output and motor performance</td>
<td>Reciprocal Imitation Training (RIT)</td>
<td>Increase in social responsivity (joint attention)</td>
<td>No improvements in motor skills</td>
<td>Preponderant</td>
</tr>
<tr>
<td>Ingersoll and Schreibman (2006)</td>
<td>M: 3, F: 2</td>
<td>Increase participants’ language output and motor performance</td>
<td>Reciprocal Imitation Training (RIT)</td>
<td>Increased joint attention, pretend play, and language (spontaneous and imitated) with therapist and caregiver</td>
<td>Increased motor imitation and spontaneous object imitation</td>
<td>Preponderant</td>
</tr>
<tr>
<td>Ketcheson, Hauk, and Ulrich (2017)</td>
<td>M: 15, F: 5</td>
<td>Increase participants’ language output, socialization, and motor performance</td>
<td>Classroom Pivotal Response Training</td>
<td>Decreased minutes in solitary time (POPE), approached sig for proximity and parallel awareness. No change in joint engagement, parallel play and onlooking</td>
<td>Increased locomotor, object control and gross quotient TGMD-2. No physical activity differences</td>
<td>Preponderant</td>
</tr>
<tr>
<td>Schaal, Benevides, Mailloux, Faller, Hunt, et al. (2014)</td>
<td>M: 17, F: 15</td>
<td>Evaluate efficacy of OT/SI following a manualized protocol on individual goal attainment</td>
<td>Occupational therapy using sensory integration</td>
<td>Decreased reliance on caregiver assistance on social subscale of the PEDI</td>
<td>Decreased reliance on caregiver assistance on Self-care subscale of the PEDI, no change in mobility subscales</td>
<td>Preponderant</td>
</tr>
<tr>
<td>Caputo, Ippolito, Mazotta, Senenza, Muzio, et al. (2018)</td>
<td>M: 17, F: 9</td>
<td>Increase participants’ language output and motor performance</td>
<td>Aquatic therapy</td>
<td>Increased on VABS Subscales of Social Abilities; CARS = emotional response, adaptation to change, activity level</td>
<td>Improved VABS – daily living skills (as did control group); Swimming skills increased, but no control comparison possible</td>
<td>Suggestive</td>
</tr>
<tr>
<td>Ingersoll and Lalonde (2010)</td>
<td>M: 3, F: 1</td>
<td>Increase participants’ language output and motor performance</td>
<td>Reciprocal Imitation Training (RIT)</td>
<td>Increase in number of word combinations and spontaneous/flexible language</td>
<td>No motor outcomes measured</td>
<td>Suggestive</td>
</tr>
<tr>
<td>Lau (2017)</td>
<td>M: 8, F: 1</td>
<td>Increase parent participation</td>
<td>Transdisciplinary home-based treatment</td>
<td>No significant change on the PEP-3</td>
<td>No significant change on the PEP-3</td>
<td>Suggestive</td>
</tr>
<tr>
<td>Miltenerger and Charlop (2014)</td>
<td>M: 3, F: 1</td>
<td>Increase participants’ motor performance</td>
<td>Athletic group games</td>
<td>Increased number of single words used communicatively, and increased number of word combination or sentences used communicatively</td>
<td>Increased locomotor, object control and gross quotient TGMD-2. No physical activity differences</td>
<td>Suggestive</td>
</tr>
<tr>
<td>Pfeiffer, Koenig, Kinnealey, Sheppard, Henderson (2011)</td>
<td>M: 32, F: 5</td>
<td>Address sensory integration intervention</td>
<td>Sensory integration therapy</td>
<td>Increase in social responsivity</td>
<td>No changes in motor performance; decrease in atypical behaviors</td>
<td>Suggestive</td>
</tr>
<tr>
<td>Preis and McKenna (2014)</td>
<td>M: 4, F: 0</td>
<td>Increase in participants’ language output</td>
<td>Sensory integration therapy</td>
<td>Increased number of single words used communicatively, and increases in spontaneous/flexible language</td>
<td>No motor outcomes measured</td>
<td>Suggestive</td>
</tr>
<tr>
<td>Yoo and Kim (2018)</td>
<td>M: 33, F: 19</td>
<td>Increase participants’ language output and motor performance</td>
<td>Dyadic drum playing</td>
<td>Increase in social skills on the K-SSRS, increase in eye gaze from pre- to post intervention; no significant change in social imitation skills</td>
<td>Increased engaged joint action; no significant decrease in asynchronizaton errors due to drum tapping</td>
<td>Suggestive</td>
</tr>
<tr>
<td>Zanobini and Solari (2019)</td>
<td>M: 19, F: 6</td>
<td>Increase in language, decrease in stereotyped behaviors</td>
<td>Aquatic Therapy</td>
<td>Increased social responsivity on SRS; Increased Social and Self-Help Skills, Sensory, Body Subscale of ABC; No significant increases on Language and Communication Subscale of ABC</td>
<td>Swimming skills increased, but no control comparison possible</td>
<td>Suggestive</td>
</tr>
</tbody>
</table>

trolled trials [12-16]. This design compared randomly assigned participants using an experimental group and a control group and the coders were blinded from the purpose of the study. Three studies had observational studies with controls [17-19]. These studies compared two groups that were not randomly assigned using an experimental group and a control group. Three studies implemented an observational study [20-22]. These studies did not compare two groups but included single case studies that monitored TF and reported valid outcomes. Two studies had an observational study without controls design [23]. These studies compared two groups that were randomly assigned; however, they did not monitor TF or use reliable and valid outcome measures. One study implemented a strong single subject design [24]. This study used a single case experimental design that monitors TF and presents valid outcome measures. There was also one study that showed a high-quality randomized controlled trial [25]. This study randomly compared two groups, used blind coders, and monitored TF.

**Treatment fidelity**

Treatment fidelity (TF) means the overall consistency of the interventions at following their outlined procedure. TF is an important indicator of internal validity. Only one study reported TF [18]. Three of the studies took TF but only for 10% of the sessions that were completed [13,24,25]. One study took measurements for TF but did not report their findings [20].

**Interrater agreement**

Interrater agreement (IRR) is the percentage of agreement between two or more observers of the treatment intervention or treatment outcomes. An IRR of 80% or greater is acceptable and having independent and blind observers was needed for the highest possible ranking. IRR is used as an indicator of the reliability of the measurements, and because of this is also used for internal validity. There were only two articles that did not report IRR [15,23]. All thirteen articles that reported IRR reached at least 80% agreement.

**Quality study appraisal**

Fifteen studies met the inclusion criteria, the quality of the individual studies varied. No studies met the criteria to be classified as conclusive. Four studies [13,18,24,25] were classified as preponderant. Nine included studies were ranked as suggestive [12,14,16,17,19-22,26]. The remaining two studies were classified as inconclusive [15,23].

**DISCUSSION**

The overarching aim of this systematic review was to determine whether motor-based interventions could lead to improved language outcomes in children with ASD. The results of this investigation were consistent with the increasing body of research that described a complex relationship between development in motor and communication skills in these children [9,10,27]. Although some of the specific language and motor results were mixed, 12 of the 13 included studies reported beneficial language outcomes in children with ASD who participated in motor-based interventions. Previously, motor function in infants and toddlers with ASD has been shown to be predictive of later communication skills [6-8]. The results of this systematic review hint at the possibility that this predictive relationship could be exploited as children...
with ASD continue to grow—that targeting motor skills in isolation or alongside language could subsequently lead to improved language outcomes. As LeBarton and Landa [6] initially posited, the inclusion of motor interventions may lessen the social communication or language delays characteristic of ASD.

Although the relationship between these two domains is still not thoroughly understood, this study indicates that the traditionally independent provision of communication and motor interventions should be reconsidered. The majority of the included studies were completed relatively recently and indicate improved language outcomes for several different motor-based interventions. Undoubtedly, co-treatment of children with ASD by occupational therapists, physical therapists, and speech-language pathologists in clinical and educational settings is occurring. However, the decision to deliver services in this form is likely influenced by practical concerns such as scheduling, proximity, convenience, and reimbursement. Evidence-based practice also requires such clinical decisions to be based on monitoring and incorporating new and high-quality research [28]. The results of the current study help justify potentially prioritizing the collaboration of clinicians in the provision of therapeutic interventions for children with ASD, even if the identified delays would have historically only supported a certain level of service.

One type of service model for coordination of therapeutic services described by Sylvester, Ogletree, and Lunnen is interdisciplinary collaborative practice (IPCP), which includes “continuous interaction and knowledge-sharing” [29] between professionals, patients, and stakeholders. The authors argue that IPCP helps advance the everyday outcomes for persons with severe disabilities (not specifically ASD), but they also acknowledge that evidence supporting collaborative processes is limited, even though position statements from both the American Physical Therapy Association and the American Speech-Language-Hearing Association endorse it [30,31]. While the findings of the current study do not directly address a specific model of collaborative service delivery, the findings indicate that measuring language outcomes is an important component of gauging the overall success of motor interventions for children with ASD, which, at minimum, necessitates the coordination of physical therapists and speech-language pathologists for clinical decision making about specific interventions and reporting progress.

Additionally, incorporating evidence-based practice into clinical decision making requires speech-language pathologists to recognize the needs, values, and preferences of individuals and families who are being provided services [28]. Parents of children who go on to be diagnosed with ASD frequently report that delays in language development are their primary concern [32]. However, parents who report concerns about motor skills in children later diagnosed with ASD were first concerned at a younger age than parents who report concerns with communication skills [33]. The same group of parents with motor concerns also sought out services sooner, but only made up a minority (approximately 23%) of the overall sample. These studies highlight the need for increased awareness of potential red flags related to motor development and ASD for both families and clinicians. The findings of the current study extend that need for increased awareness into the area of intervention. As previously noted, LeBarton and Landa [6] suggested that earlier provision of motor interventions may help alleviate the influence of motor delays on later communication skills in children with ASD. If families were more informed about the potentially facilitative influence of motor interventions on language outcomes, their preferences and values could be more specifically incorporated.

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REFERENCES


