International Journal of Speech-Language Pathology, 2014; 16(1): 1–10

informa
healthcare

SCIENTIFIC FORUM: LEAD ARTICLE

Early identification and early intervention in autism spectrum disorders: Accurate and effective?

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Abstract
Over the past decade, there has been increased interest in identifying autism and autism spectrum disorder (ASD) in toddlers. Although there is a strong rationale for identifying ASD early and delivering effective intervention, a recent report in the journal *Pediatrics* raises important questions about the scientific evidence currently available supporting early intervention. In addition, the British National Health Service (NHS) has not adopted universal screening for autism, even though the American (US) Academy of Pediatrics endorsed a recommendation that all toddlers be screened for ASD by the age of 24 months (in 2007). The goal of this initiative is to identify and, where indicated, provide early intervention for autism and ASD. Although it is inarguable that this is a worthwhile and laudable goal, the systematic study of this goal is confounded by the inherent difficulty in reliably identifying autism in 24-month-old toddlers. It is challenging to demonstrate intervention effects in the absence of randomly assigned control groups in an increasingly heterogeneous ASD population. The purpose of this paper is to examine the current literature on early identification and early intervention in autism and ASD and to provide a framework for examining these issues.

Keywords: *Autism spectrum disorder, early identification, early intervention, autism spectrum disorder screening, autism spectrum disorder treatment.*

Introduction and background

Warren, McPheeters, Sathe, Foss-Feig, Glasser, and Veenstra-VanderWeele (2011) reported that there is currently relatively weak evidence in support of the effects of early intervention in treatment of ASD. This conclusion was based on a review of the evidence in studies in autism and autism spectrum disorders and appeared in *Pediatrics*, a flagship medical journal in the US. How could this possibly be the current state of the art? After all, there is a high degree of face validity to the notion that identifying and treating autism in young children will improve outcomes. Also, there have literally been hundreds of studies on treating the various aspects of autism and many have yielded positive results (see Koegel, Koegel, & Camarata, 2010). However, the Warren et al. review highlights the difference between demonstrating the effects of a particular treatment on autism symptomology and objectively testing early intervention. Although the implications of the current paucity of evidence extend to all clinicians delivering early intervention for autism, this issue also has profound direct implications for speech-language pathologists. The purpose of this review is to examine the factors that potentially impact the evidence base for early identification of autism and testing early intervention. It is possible that the current weak evidence in support for early intervention is rooted in the evolution of the diagnosis and identification of autism (and subsequently the autism spectrum) and ongoing evolution for diagnostic criteria.

"Infantile Autism" was first classified by Kanner (1943) and for nearly 50 years was viewed as a relatively rare, low incidence condition. From the beginning, diagnosis and treatment focused on addressing the three core symptoms that coalesced to produce the unique features of autism: (a) reduced motivation for social interaction, (b) restricted interests and repetitive behaviours, and (c) severe communication disorders. Originally, disruptions in *all three domains* were required for diagnosis. More recently, following the reconceptualization of autism as a broader "spectrum" disorder, the reported incidence rate has increased dramatically (US Centers for Disease Control, 2012). Of particular note in this evolution was the modification of the diagnostic criterion on the reduced motivation for social interaction. In the Diagnostic and Statistical Manual of Mental Disorders 3rd edition (DSM-III;

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ISSN 1754-9507 print/ISSN 1754-9515 online
Published by Informa UK, Ltd.
DOI: 10.3109/17549507.2013.858773

APA, 1980, p. 89), this was a "pervasive lack of responsiveness to other people". In the subsequent edition (DSM-IV-TR, 1994), "qualitative impairment in social interaction" had replaced "pervasive lack of responsiveness to other people" (APA, 1994, p. 70).

This evolution from "infantile autism" to "autism spectrum disorder" (ASD) has also been an ongoing source of confusion among clinicians and parents. Broadly, the field has been struggling with what constitutes the boundaries of autism and there are ongoing controversies on precisely what constitutes autism and autism spectrum disorder (Swedo, Baird, Cook, Happé, Harris, Kaufmann, et al., 2012). Autism or autistic disorder (AD) is a more severe and often more persistent form of the disorder relative to subtypes (spectrum) that do not include all three autistic traits. That is, it is clear that children who display a sub-set of autism symptoms rather than the full pantheon of reduced social motivation, communication disorders, and restricted interest that are the hallmarks of the condition do not develop in the same way as those who simultaneously display all or multiple autism traits. It is not surprising that the popular press and, in some cases, the scientific literature conflates AD and ASD. However, as we will see, this distinction is very important because unbiased study of early identification and early intervention requires reliable and valid diagnosis. Moreover, the current epidemiological data suggest important differences in the reliability and validity for identifying autism (AD) as compared to other ASD subtypes (Rondeau, Klein, Masse, Bodeau, Cohen, & Guilé, 2011). Indeed, the proposed revisions in the DSM-5 have excluded two ASD sub-types previously listed in the DSM-IV, due, at least in part, to the instability and lack of concordance for these sub-types (Lord & Jones, 2012).

From an early identification and early intervention perspective, it is also noteworthy that early-onset was prominent in Kanner's original characterization of the condition, including the specific contention that these core symptoms were evident from the beginning of life, that is, in infancy. Indeed, the term "infantile autism" was used in the DSM-III, which was the first edition to specifically include autism (earlier editions referred to "childhood schizophrenia"). However, in clinical practice, autism was often first diagnosed in early school-age or even older children, and toddler or pre-schooler age diagnoses were much rarer. Because of this, there have been ongoing efforts to develop more specific nosology and objective measures to capture the symptomology at earlier and earlier ages (Lord & Jones, 2012; Volkmar, Cohen, & Paul, 1986), including the development and refinement of the Autism Diagnostic Observation Scale (ADOS; Lord, Risi, Lambrecht, Cook, Leventhal, DiLavore, et al., 2000; ADOS-2; Lord, Rutter, DiLavore, Risi, Gotham, & Bishop, 2012). This, in turn, has led to more systematic diagnostic practice focused on early identification, and served as a psychometric platform for developing tools for increasingly earlier diagnosis, with the ultimate goal of generating accurate diagnosis in toddlers and infants.

One of the most important motivations for developing more accurate and earlier identification protocols has been the long-range goal of ameliorating the symptomology by developing and delivering effective early intervention. If one adopts the perspective that autism is, in fact, a lifelong disabling condition with an ontology in infancy (see Koegel & Koegel, 2006), there is a high degree of face validity to the notion that early intervention can improve outcomes relative to intervention that is delivered later in life. This perspective is particularly salient in autism because the reduced motivation for social interaction coupled with the over-selectivity that results in restricted interests, children with untreated autism are especially likely to miss important learning cues from parents, siblings, and peers. That is, if the motivation for social communication were increased and the over-selectivity, at least to the extent where this autism trait interferes with processing important learning cues, were decreased, it is inarguable that effective early intervention would result in improved outcomes. Although a similar argument could be made for nearly any developmental disability, including speech and language disorders, the broadly pervasive, intractable, and persistent deficits seen in autism lend a particular urgency to these efforts in this population.

Thus, there is a compelling pedagogy for delivering early intervention that is based upon accurate early identification. Given this backdrop, it is perhaps unsurprising that, in 2007, the American Academy of Pediatrics issued a policy statement calling for universal screening for autism by the age of 24 months (Johnson & Myers, 2007). What is perhaps more surprising is the ongoing recommendation *against* universal screening by the National Health Service (NHS) in the UK (http://www.screening.nhs.uk/autism, Allaby & Sharma, 2011). At the risk of over-simplification, the key difference in the Academy of Pediatrics position and the NHS position arises from the practical difficulties associated with accurately identifying autism specifically or ASD more broadly in toddlers (24-month-olds). Allaby and Sharma argue the current ability to correctly detect ASD (sensitivity) and differentially diagnose the condition (specificity) continues to be problematic in toddlers and pre-schoolers so that nationwide implementation in the UK is not feasible. In contrast, the Academy of Pediatrics recommends general screening procedures followed by a comprehensive evaluation in toddlers by the age of 2 years. Although not explicitly stated in the Academy of Pediatrics position paper (Johnson & Myers 2007), the impetus for this initiative is rooted in delivering early intervention.

The issue of early identification and early intervention for autism and ASD is important for speech-language pathologists because many of the core traits associated with this disability relate to speech and language development. Moreover, speech-language pathologists are likely to be directly or indirectly involved in both the initial differential diagnosis and in the intervention process. Indeed, as Johnson & Myers (2007) note: "Most children who are later diagnosed with AD and PPD-NOS present to their PCP [primary care physician] with 'speech delay'" (p. 1191). Stated simply, the initial referral is overwhelmingly based upon the late onset of talking, which is what catches the attention of parents, paediatricians, and family physicians. However, this diagnostic marker, although salient in AD and ASD such as Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), is not specific to these conditions so that differential diagnosis of speech sound disorder, language disorder, and global intellectual disabilities (as well as hearing loss) becomes paramount. Although this would seem to be relatively straightforward, the truth is that accurately performing this kind of differential diagnosis, which is crucial for accurate identification and ultimately for developing and testing early intervention, can be difficult in toddlers. Heterogeneity and difficulty in constructing objective testing procedures in this population increases reliance on observational scales and subjective judgements. A review of the historical background and core traits of autism and the ongoing development of the criteria and procedures for identifying autism spectrum disorder may be helpful in clarifying the key traits that differentiate autism from communication disorders.

Diagnosing autism and autism spectrum disorder: A historical perspective

There is nearly unanimous consensus among clinicians and scientists that autism is a lifelong severe disabling condition with primary deleterious effects on social engagement, rigid routine preferring behaviour patterns and unusual behavioural traits such as stereotypy, and, in some cases, self-injury (Lord et al., 2012). Of particular importance to speech-language pathologists are the disruptions in communication development. The Diagnostic and Statistical Manual of the American Psychiatric Association (2013) indicates that "the essential features of autistic disorder are the presence of markedly abnormal or impaired development in social interaction and communication and a markedly restricted repertoire of activity and interest." (p. 66). Similarly, in order to be diagnosed with any Pervasive Developmental Disorder, which is the superordinate classification for Autism, Asperger Syndrome, Childhood Disintegrative Disorder, Rhett's Disorder, and PDD-NOS, a child must display: "severe and pervasive impairment in several areas of development: reciprocal social interaction skills, communication skills, or the presence of stereotyped behaviour, interests, and activities" (p. 65) DSM-IV APA 1994. Collectively, several Pervasive Developmental Disorder typologies, including Autism, Asperger Syndrome, and PDD-NOS, have come to be known as Autism Spectrum Disorder (ASD; Johnson & Myers, 2007). Further, the DSM-IVR indicates, "the qualitative impairments that define these conditions are distinctly deviant relative to the individual's developmental level or mental age" (p. 65).

Originally, Kanner (1943) noted the pervasive differences in some of his patients with intellectual disabilities in the children's psychiatric service at Johns Hopkins University. The primary trait distinguishing these patients was a markedly reduced motivation for social interaction which was noteworthy in the broader population of patients with intellectual disabilities, who, despite significantly reduced intellectual abilities, continued to display relatively higher levels of social engagement and social mobility than their counterparts with autism. He noted "we must assume that these children have come into the world with an innate *inability* to form the usual, biologically provided affective contact with people" (Kanner, 1965, p. 412), and also argued that the symptomology was evident from birth. Moreover, it was clear that Kanner conceptualized autism as a specific confluence of key symptoms with a lack of social attachment as the centrepiece of the symptomology. He also recognized and criticized the tendency to dilute the symptomology or view autism from a piecemeal perspective. In 1965 he noted

> While the majority of the Europeans were satisfied with a sharp delineation of infantile autism as an illness *sui generis* [a thing unto itself, unique], there was a tendency in this country [US] to view it as a developmental anomaly described exclusively to maternal emotional determinants. **Moreover, it became a habit to dilute the original concept of infantile autism by diagnosing it in many disparate conditions, which show one, or another isolated symptom found as a part of the feature of the overall syndrome. Almost overnight, the country seemed populated by a multitude of autistic children, and somehow this trend became noticeable overseas as well. Mentally defective children who displayed bizarre behaviour were promptly labelled autistic …** (p. 413) [emphasis added].

Dr Kanner further noted:

> By 1953, van Krevelen rightly became impatient with the confused and confusing use of the term infantile autism as a slogan indiscriminately applied with cavalier abandonment of the criteria outlined rather succinctly and unmistakably from the beginning. He warned against the prevailing "abuse of the diagnosis of autism," declaring that it "threatens to become a

> fashion." A little slower to anger, I waited until 1957 before I made a similar plea for the acknowledgment of the specificity of the illness and for adherence to the established criteria (p. 413).

It is clear that professor Kanner wished to be specific regarding the diagnosis of autism. Importantly, for the purposes of this review, Kanner's parsimony was evidently rooted in ensuring that these people received treatment that was properly focused on autism symptomology.

To be sure, advances in psychometrics and advances in identifying autism have been made since Kanner's original description and his subsequent exhortations for precision in diagnostic practice. On the other hand, there may be pitfalls with regard to Kanner's concerns as procedures for identifying autism and the broader autism spectrum are applied to younger and younger children. In particular, it is often the persistence of normal behaviour far beyond the bounds of what is consistent with typical development that distinguishes autism. For example, many typically-developing toddlers move their hands in repetitive, stereotypical patterns that would be unusual, and perhaps even symptomatic of autism in a 4- or 5-year-old child. The ubiquity of this behaviour in typically-developing toddlers reduces its utility for autism screening in 2-year-olds (see Turner & Stone, 2007). This is an important point: a diagnostic marker for autism in a 4- or 5-year-old may not be useful in a 2-year-old precisely because it is the persistence of the behaviour beyond the toddler period that is so salient in autism.

From an early intervention perspective, this is crucial. Assume that a language-impaired toddler is mistakenly identified as having PDD-NOS (and thus ASD) and is enrolled in an early intervention study. Further assume that one of the "symptoms" of autism is repetitive behaviour that in fact is simply normal variation in typical development. When this "typical" repetitive behaviour diminishes simply because of maturation, the clinical scientist may mistakenly conclude that the reduced repetitive behaviour is in fact evidence that the early intervention was effective. Conclusive evidence of the effectiveness of early intervention must take into consideration normal variation and untreated maturation (spontaneous recovery). Another way of making this point is to say that therapeutically-induced reductions of stereotypy in a 6-year-old may be easier to substantiate from an intervention perspective because the repetitive movements often seen in typical toddlers have substantially disappeared by this time. Therefore, taxonomies of ASD symptoms and the specific traits that are targeted in early intervention must include controls for typical development and confounding factors in other, non-ASD, clinical populations such as speech disorder, language disorder, and global intellectual disabilities. This is especially important for non-ASD communication disorders in toddlers that may initially have substantial overlap with autism.

Thus, although speech-language pathologists typically do not serve a lead or primary role when diagnosing autism and other ASD conditions, the core symptomology relating to communication disorders, pragmatic (social) skills, and other speech and language related abilities suggests that this profession should be a foundational member of the diagnostic and treatment team. Because of this, it is important that speech-language pathologists serving children with autism and other forms of ASD be well versed in the diagnostic, and, perhaps more importantly, the evidence base for social, speech, and language intervention in these children. The increased focus on early intervention often results in identification of autism or another form of ASD at age 2 years or even earlier, wherein the primary symptom generating the autism or ASD referral is late onset of speech and/or language (Johnson & Myers, 2007). Thus, initial early differential diagnosis of speech and/or language disorder by a speech-language pathologist is a crucial aspect of the assessment process for children with PDD-autism or other forms of ASD.

Historically, in order to be diagnosed with autism, a child must have displayed reduced social attachment (and associated reductions in social skills), in addition to a noteworthy degree of affinity for sameness and routine. Although it is certainly true that Kanner's (1943) original description asserted that autistic symptomology is present from the beginning of life; until the past decade, the majority of diagnoses were in children, often at least 4 or 5 years of age, rather than toddlers and infants. Also, delayed language unto itself was often less prominent in these older children than the persistence of tantrums, social detachment, and unusual behaviours that are the hallmarks of PDD-Autism. It is noteworthy that the DSM-IV-TR and the proposed guidelines for the DSM-5 include a separate diagnostic classification for communication disorders such as language disorder so that differential diagnosis of autism and other ASDs explicitly requires symptomology beyond what can be accounted for by language disorder or speech sound disorder alone. In 5-year-olds, this is relatively straightforward and identifying autism at this age has a high degree of both sensitivity and specificity (see Lord et al., 2000). In contrast, differential diagnosis in infants and toddlers can be much more challenging and particularly difficult in cases of language disorder wherein there are significant deficits in auditory comprehension (Mixed Expressive-Receptive Language Disorder in the APA 2000 DSM-IV-TR).

Unfortunately, this high degree of accuracy, in terms of sensitivity and specificity, for an autism diagnosis with relatively severely affected children at an older age comes with a potential cost: there

has likely been a lost opportunity for effective early intervention that may plausibly have reduced the symptomology of ASD. Therefore, the past decade has seen a call for early identification and treatment for autism and the other forms of ASD in 2-year-old pre-school children (for example, see Zwaigenbaum, Thurm, Stone, Baranek, Bryson, Iverson, et al. 2007). This effort resulted, in 2007, in the American Academy of Pediatrics (Johnson & Myers, 2007) publishing guidelines on the early identification of children with autism spectrum disorder and recommending universal screening of all toddlers in the US by the age of 24 months. As mentioned previously, the National Health Service in the UK has not followed suit and, to date, does not recommend universal screening for ASD in 2-year-old children (http://www.screening.nhs.uk/autism, Allaby & Sharma, 2011). To be sure, this does not mean that the NHS is opposed to early identification and treatment for autism, rather, this position is evidently rooted in the notion that sufficient evidence must accrue prior to adopting and implementing universal screening for autism prior to the age of 5 years.

Clinical scientists and practicing speech-language pathologists may be surprised to learn that there is any question of whether screening toddlers and young pre-schoolers for ASD should be completed. After all, early identification and early intervention is directly predicated upon the ability to reliably recognize, and subsequently treat symptoms of autism and other forms of ASD, and there appears to be consensus, at least in the US, that this should be completed for toddlers before the age of 2 years. Indeed, from a theoretical perspective, how could anybody be opposed to early identification and early intervention for autism or other forms of ASD?

A review of the current evidence on early identification and early intervention, with a particular focus on the symptomology most relevant to speech-language pathologists, may be instructive. A general framework for examining evidence needed to support early intervention will be provided. In addition, the current gaps in this evidence will be discussed. Of particular interest to speech-language pathologists is the importance and relevance of the emergence of first words, especially as this relates to social language disorders (pragmatics) as a marker for autism and other forms of ASD as well as for language disorder.

Stability of AD as compared to ASD: Implications for early intervention

Starting from the earliest efforts to distinguish autism from other disability typologies, one of the most important markers for autism is a reduced motivation for both verbal and non-verbal social engagement (see Kanner, 1943). In practice, because social interaction is such a foundational characteristic in many children, this symptom turns out to be relatively salient: parents, clinicians, and teachers are often readily able to detect severe disruptions in social abilities. Additionally, the literature on long-term stability of "full" autism (PDD-Autism, also called "Kanner's Autism" or "classic" autism) indicates a high degree of concordance between early identification and subsequent diagnostic classification. For example, Rondeau et al. (2011) reported a 76% stability rate for an autism diagnosis prior to the age of 3 years in a meta-analysis of studies examining long-term follow-up in children originally diagnosed with autism or a different form of ASD: PDD-NOS. In contrast to those initially diagnosed with classic autism, the PDD-NOS group had a long-term stability of 35%. Rondeau et al. concluded:

> the meta-analysis confirms the hypothesis that there is a higher diagnostic stability for AD than PDD-NOS. Pooling data from the selected studies indicated that an AD [Autistic Disorder] diagnosis was stable over time (76% stability) whereas PDD-NOS tended to be unstable over time (35% stability) (p. 4).

Although there are multiple potential reasons for these very different stability rates in classic autism as compared to PDD-NOS, Rondeau et al. (2011) speculated that, among other factors, a developmental disorder, such as communication disorder, that became less severe over time, accounted, at least in part, for the lower stability of the ASD that was not classic autism or "full" autism. It is clear that those children presenting with full autism symptomology, especially displaying noticeably reduced verbal and non-verbal social engagement, are relatively easy to identify at an early age, and that the long-term stability for this early identification is relatively high. However, there is far less diagnostic stability over time for the children who do not display full autism symptomology and are placed on the "Autism Spectrum" based on PDD-NOS diagnosis. Clearly, testing the effectiveness of early intervention requires accurate early identification. At this time, it is safe to say that this can be done more readily in AD but would be more problematic in the PDD-NOS form of ASD. It is also clear that conflating or pooling AD and PDD-NOS into an "ASD" treatment group will likely yield high variability, low stability, and potentially uninterpretable or inconclusive results.

Central role of speech-language pathologists in early identification

Because the initial identification for autism is often completed by child psychiatrists, developmental paediatricians, and/or clinical psychologists who may have relatively limited training and experience in toddlers and pre-schoolers with language disorders or other communication disorders, speech-language pathologists potentially have an important role in the differential diagnosis of toddlers and pre-schoolers who do not display full or classic

autism symptomology. From this perspective, it may be useful to review conditions in the Diagnostic and Statistical Manual that include late onset of words as a possible diagnostic classification.

In addition to PDD-Autism and the other forms of ASD, late onset of words can also be a diagnostic marker for mental retardation/intellectual disabilities, language disorder, and/or speech sound disorder. As Camarata and Nelson (2002) pointed out, diagnostic instruments such as the Autism Diagnostic Observation Schedule (ADOS), although useful for identifying core traits in the language and social aspects of autism, may not be well suited to differential diagnosis of ASD, language disorder, and/or speech sound disorder. Receptive language disorder (mixed expressive and receptive language disorder in the DSM scheme) is likely to be particularly problematic, especially in toddlers and pre-schoolers. From a theoretical perspective, the key diagnostic marker separating ASD from receptive language disorder should be non-verbal social engagement: children with ASD should display reduced social motivation exemplified by low verbal and non-verbal social engagement. In contrast, although children with receptive language disorder will, like those with ASD, display reduced verbal social engagement arising from poor communication skills, presumably the non-verbal social engagement should be relatively preserved. That is, differential diagnoses rest squarely upon the degree to which each group displays non-verbal social engagement. The key markers for non-verbal engagement in toddlers include initiating and responding to joint attention, reciprocal social smile, and displaying shared enjoyment (see Lord & Risi, 2000; Lord et al., 2012; Stone & Yoder, 2001; Wetherby, Woods, Allen, Cleary, Dickinson, & Lord, 2004), with these aspects significantly diminished in toddlers with autism and much less so in those with receptive language disorder.

Another important consideration is that many children with late onset of word production evidently are displaying a variation on typical development rather than clinical symptomology (see Camarata, 2012; Rescorla & Dale, 2012). That is, a number of studies have suggested that there is a relatively high spontaneous recovery rate for toddlers at the age of 24 months, provided the late onset of words is the sole symptomology (see Ellis-Weismer, 2007; Rescorla, 2002; Whitehurst, Fischel, Lonigan, Valdez-Menchaca, Arnold, & Smith 1991; Whitehurst, Fischel, Arnold, & Lonigan, 1992). This is noteworthy because the "spontaneous recovery" phase for these "late bloomers" is almost exclusively before the age of 5 years, which coincides with the age (5 years and older) at which there is very high stability in diagnosis of autism and other forms of ASD. Stated simply, it is an interesting question as to whether the reduced long-term stability in identifying PDD-NOS (which is the overwhelmingly highest category of ASD identified in toddlers and pre-schoolers) is associated with the developmental course seen in "late bloomers" and clinical populations of children with speech sound disorders or language disorders who are initially classified as ASD.

This orientation on the inherent challenges associated with stability in early diagnosis of classic autism as compared to other forms of ASD such as PDD-NOS provides a framework for discussing evidence needed to test whether early intervention is effective. That is, a key element in establishing credible evidence for early intervention is (a) accurate and stable diagnoses and, more importantly, (b) evidence that early intervention produces gains that are greater than spontaneous recovery rates. In classic autism, the severity of the symptomology yields relatively stable diagnostic classification, even in toddlers and pre-schoolers. In contrast, the other forms of ASD, such as PDD-NOS, do not currently display this level of stability.

Current evidence to support early intervention

Clinical scientists have long recognized that establishing credible evidence for a treatment effect is not necessarily a straightforward endeavour. One of the most difficult challenges is that many illnesses have the baseline rate of growth or spontaneous recovery in toddlers and pre-schoolers. A recent review by Warren et al. (2011) of the literature on early intervention in ASD describes the key features that are needed for a credible study of the impact of early intervention on toddlers with ASD. These include minimizing the risk of bias, using a reasonable study design (such as a randomized controlled trial), employing a credible diagnostic approach including a standardized autism diagnostic instrument combined with clinical judgement, having adequate descriptions of participant characteristics, comprehensive intervention descriptions (including treatment fidelity), relevant and plausible outcome measures that are directly relevant to autism symptomology, and a proper statistical approach to analysing the data. In particular, protecting against bias (e.g., study blinding), accurate diagnosis, and proper design are crucial for establishing credible evidence that early intervention has a positive impact.

Interestingly, an ongoing challenge in the intervention literature is that many studies evidently are designed to be either implicitly or explicitly *confirmatory* for a particular approach rather than as a *randomized fair test* of the intervention. For example, in Lovaas (1987), the participants were not randomly assigned to the two comparison conditions, rather those that responded to imitative prompts were assigned to the condition that ultimately proved to be superior, whereas those that would not imitate during pre-test were assigned to the comparison condition. It would be interesting to know whether participants who did not readily respond to imitative prompts would have made similar gains (see Gillum,

Camarata, Nelson, Camarata, 2003). Similarly, including heterogeneous participants with unstable diagnoses makes it difficult to reliably detect differences between comparison groups. That is, if an ASD sample includes a relatively high proportion of the less stable PDD-NOS sub-type, this variability may obscure important main effects for the early intervention.

Given this state of affairs, it is perhaps not surprising that the results of the comprehensive review by Warren et al. (2011) indicated: "The strength of the evidence [to support early intervention] overall ranged from insufficient to low" (p. 1303). It is noteworthy that this review appeared in high-impact journal *Pediatrics*, so there is a pressing need for fair and objective studies of early intervention. To be sure, this comprehensive review suggested a number of promising findings such as:

> Studies of Lovaas-based approaches and early intensive behavioural intervention variants and the Early Start Denver Model resulted in some improvements in cognitive performance, language skills, and adaptive behaviour skills in some young children with ASDs, although the literature is limited by methodologic concerns (Warren et al., 2011),

and "Data suggest that sub-groups of children displayed more prominent gains across studies, but participant characteristics associated with greater gains are not well understood" (Warren et al., 2011, p. 1303). This latter conclusion is intriguing because it calls for more specificity in identifying individual child characteristics and matching these to the specific intervention procedures. That is, from a theoretical perspective, if one has a generic pool of toddlers with ASD who display variable autism symptomology, and intervention provided is designed to treat *all* autism symptoms or a sub-set of symptoms that does not match a significant portion of the participant pool, it is hardly surprising that such a study ultimately yields weak evidence. On the other hand, when the autism symptomology is precise and the interventions match the child's traits, one could plausibly hypothesize that such a study would be more likely to yield interpretable results.

Receptive and expressive language deficits as a core trait of autism and ASD

As mentioned earlier, a core trait of Autism and ASD is severe disruptions in language development, limiting the extent children can participate in social interaction (Bruinsma, Koegel, & Koegel, 2004; Haines & Camarata, 2004; Koegel & Koegel, 2006; Marcus, Garfinkle, & Wolery, 2001). Because language skills are among those most disrupted, even relative to other abilities in children with ASD, improving *receptive* and expressive language often has a high priority for early intervention. Not surprisingly, verbal abilities and programs targeting improved verbal skills have long been a primary focus in the treatment of ASD (e.g., Lovaas, 1971; Lovaas, Schriebman, & Koegel, 1974; Smith & Camarata, 1999). Moreover, simultaneous receptive and expressive language deficits are one of the defining characteristics of both AD (PDD-Autism) and PDD-NOS. Note that one form of ASD, Asperger Syndrome, includes typical syntactic and lexical skills.

Language ability is generally considered to include *both* comprehension and production (Lahey, 1988) and competence in both domains is required for successful educational development and also for social interaction. Without minimizing the importance of language production, auditory language comprehension also has a key role in typical development and in disabling conditions such as ASD (Gillum & Camarata, 2004). It is, thus, noteworthy that, in contrast to language production, very few studies have examined whether language *comprehension* (receptive language) can be improved in ASD and whether receptive language will improve as a consequence of expressive language intervention (as appears to be a widely held assumption). For example, our recent review (Gillum & Camarata, 2004) identified numerous assessment studies indicating children with ASD *universally* display significant receptive language deficits across severity levels (e.g., "low" and "high" functioning) and there are dozens of studies examining the effects of treatment on language production (words, speech-intelligibility, and grammar, see Lord et al., 2012; Warren et al., 2011). Stated simply, ample evidence exists that nearly ALL children with ASD (except PDD-Asperger Syndrome) have severe deficits in auditory language *comprehension* as well as language production, and these deficits have profound and far-reaching impacts on social development and on access to educational opportunities. Given the inherent challenges in treating this broadly disabling condition which has wide ranging effects on family, educational institutions, access to society, and self-determination, there is a clear need to study *receptive* language abilities in ASD, especially relative to early identification and early intervention.

Experimental studies of word learning and grammar indicate children with disabilities are less skilled in each of these areas than their typically-developing peers. Like children with Down Syndrome (Chapman, Hesketh, & Kistler, 2002), children with ASD are particularly likely to display dissociations between receptive and expressive language skills, often naming objects for which they show difficulty comprehending (Camarata & Nelson, 2006). In typical development a strong inter-relationship exists between receptive and expressive language (Camarata, 2000). This interrelationship has been described by accounts of the transactional model of language acquisition and bootstrapping models. The transactional model (Camarata & Nelson, 2006; Snyder-McLean & McLean, 1987; Yoder & Warren, 1993) serves as a description of the interaction between comprehension and expression

during real-time interaction between caregiver and child (Moerk, 1992).

However, differential diagnosis of ASD and language disorder may be particularly problematic in toddlers. Both groups are likely to express frustration in the form of tantrums, and both will have impaired verbal social skills and be generally unresponsive to verbal input. Indeed, one could argue that highly accurate diagnosis can occur only after there has been an improvement in the receptive language abilities so that a clinician could explore whether autism symptomology remains after the communication disorders are reduced. Naturally, distinguishing between receptive language disorders and ASD is relatively straightforward in 5- and 6-year-olds (and older children), but much more difficult in toddlers. However, both populations are much more likely to be persistent in their symptomology than are children with early language delay, but no auditory comprehension deficits. Taken together, this suggests that early identification should include careful scrutiny of receptive language abilities and non-verbal social abilities as important diagnostic markers distinguishing language disorder from ASD. In addition, one could argue that children with language disorders without receptive deficits should be excluded from the autism spectrum *unless* additional autism symptomology in the behavioural domains is evident (Bishop & Norbury, 2002). Moreover, there is broad consensus that language disorder is not a form of ASD and should be differentially diagnosed. Lord & Jones (2012) argue:

> The argument for considering language delay as a separate dimension that is not part of ASD diagnostic criteria, but critical to how those diagnostic criteria are used, is two-fold. First, language delay is not specific to autism (Bishop & Norbury, 2002). By far the majority of young children referred for concerns about language delay *do not have ASD* or even severe developmental delays (Ellis & Thal, 2008), conversely, not all, though many children with ASD have language delays (Baird, Charman, Pickles, Chandler, Loucas, Meldrum, et al., 2008; Kjellmer, Hedvall, Fernell, Gillberg, & Norrelgen, 2012) (p. 493) (emphasis added).

In practice, it appears that the distinction between language disorder and ASD can be blurred in toddlers. For example, the recent surge in ASD eligibility reported in California (in the US) was proportionally offset by a reduction in speech and language eligibility, implying that cases were migrating from one category to another. To be sure, it is certainly possible that previously unrecognized ASD had been misidentified as language disorder. On the other hand, it is also possible that coincidental revisions of eligibility criteria for ASD coupled with the aforementioned instability of PDD-NOS contributed to the reported increase. Adding to the confusion is the inherent, but not well understood, difference in establishing eligibility for early intervention services and obtaining a DSM diagnosis. In the US, individual states have latitude in establishing criteria for early intervention eligibility, so it is possible for a child with language disorders who otherwise does not meet the DSM criteria for ASD diagnosis to be eligible for enrolment in early intervention services under special education ASD criteria. That is, the child may not receive an ASD medical *diagnosis* but be *eligible* for ASD special education services. Not surprisingly, this can lead to confusion among parents and clinicians and, from a broader perspective, make it difficult to conduct fair studies of the effectiveness of early identification and early intervention.

Future directions: A call to action

From a broad perspective, the current state-of-the-art with regard to evidence to support early identification and intervention is not as strong as many clinicians may suppose. Proposed autism guidelines in the DSM-5 (Swedo et al., 2012) may shift incidence and eligibility parameters without necessarily seeing a *real* change in the actual incidence of ASD (see report from the US Centers for Disease Control, 2012). Also, many countries currently provide early intervention services for children identified as having some form of ASD, and, in the US, increasing legislative initiatives to require private insurance companies to reimburse intervention services for ASD. Collectively, there appears to be a presumption that these early intervention services are supported by evidence. However, due to the increasingly difficult economic situation in most countries, there is increasing system-wide pressure to reduce healthcare costs (including allied health) and, in the US, pressure from private insurers to document the evidenced-based positive impact of the services as a requirement for reimbursement. Thus, there is a clear need to expand the evidence base. Indeed, this need is especially crucial, as reimbursement for healthcare services either from public or private entities is increasingly dependent upon credible scientific evidence.

Candidly, there is no doubt in my mind that toddlers with ASD can be reliably identified and that early intervention is potentially highly effective in reducing long-term ASD symptomology. However, *proving* this to an increasingly sceptical healthcare and educational environment is an altogether different matter than asserting these beliefs. Frankly, the Warren et al. (2011) review should serve as a wake-up call to clinical researchers, as the evidence base supporting early intervention is not nearly as strong as one would suppose. However, this will also require fair and objective studies and, more importantly, an unbiased approach to systematically asking the hard questions about what works and what doesn't. Testing intervention effects on the marginal cases, those who have minimal ASD symptomology,

while otherwise warranted, is not likely to lead to interpretable, credible studies. On the other hand, testing early intervention exclusively in the more severe cases, that is, those who display classic, full symptom AD, is limiting our evidence to precisely those children who are least likely to change and, thus, is a very difficult challenge.

Going forward, speech-language pathologists will play a central role in testing the validity of early identification and the impact of early intervention. Indeed, given the core and ubiquitous nature of language deficits in ASD symptomology, and the positive outcomes in these children if these can be ameliorated, one could argue that this represents an area with significant growth potential for the field, both in terms of research and practice. Of particular importance is differential diagnosis of speech sound disorder, language disorder, including receptive language disorder, and ASD in toddlers. There is also a critical need for developing effective receptive language interventions for these populations. It is perhaps small comfort that other professions and approaches are undergoing similar scrutiny. For example, a recent policy statement on the lack of evidence to support *sensory integration therapy* was issued by the American Academy of Pediatrics (2012). Sensory integration therapy is a widely used, but unsubstantiated, approach to treating ASD and other disabilities. Thus, in the near future, it will no longer be sufficient to assert the importance of early identification and early intervention simply on the basis of pedagogical arguments. It will increasingly behoove our profession to bring credible evidence to these discussions.

Declaration of interest: The author report no conflicts of interest. The author alone is responsible for the content and writing of the paper.

References

Allaby, M., & Sharma, M. (2011). *Screening for autism spectrum disorders in children below the age of 5 years: A draft report for the UK National Screening Committee*. Oxford, UK: Solutions for Public Health.

American Academy of Pediatrics. (2012). Policy statement: Sensory Integration Therapies for children with developmental and behavioral disorders. *Pediatrics*, *129*, 1186–1189.

American Psychiatric Association. (1980). *Diagnostic and statistical manual of mental disorders (III)*. Washington DC: American Psychiatric Association.

American Psychiatric Association. (1994). *Diagnostic and statistical manual of mental disorders (IV)*. Washington DC: American Psychiatric Association.

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders (IV-TR)*. Washington DC: American Psychiatric Association.

American Psychiatric Association. (2013). *Diagnostic and statistiacal manual of mental disporders (5)*. Washington, DC: American Psychiatric Association.

Baird, G., Charman, T., Pickles, A., Chandler, S., Loucas, T., Meldrum, D., et al. (2008). Regression, developmental trajectory and associated problems in disorders in the autism spectrum: The SNAP study. *Journal of Autism and Developmental Disorders*, *38*, 1827–1836.

Bishop, D. V., & Norbury, C. F. (2002). Exploring the borderlands of autistic disorder and specific language impairment: A study using standardised diagnostic instruments. *Journal of Child Psychology and Psychiatry*, *43*, 917–929.

Bruinsma, Y., Koegel, R. L., & Koegel, L. K. (2004). Joint attention and children with autism: A review of the literature. *Mental Retardation and Developmental Disabilities Research Reviews*, *10*, 169–175.

Camarata, S. (2003). Assessment of language and language disorders. In M. Wolraich (Ed.), *Disorders of development and learning* (3rd ed.) (pp. 49–60). Hamilton, Ontario: B.C. Decker.

Camarata, S. (2012). Which late talkers require intervention? Matching child characteristics and risk factors to treatment. In L. Rescorla, & P. Dale (Eds.), *Late talkers: Language development, interventions and outcomes* (pp. 303–324). Baltimore, MD: Paul H. Brookes.

Camarata, S., & Nelson, K. (2006). Conversational recast intervention with preschool and older children. In R. J. McCauley, & M. Fey (Eds.), *Treatment of language disorders in children* (pp. 237–264). Baltimore, MD: Paul H. Brookes Publishing.

Camarata, S., & Nelson, K. E. (2002). Measurement and the diagnosis of speech and language disorders in children. *Peabody Journal of Education*, *77*, 106–116.

Camarata, S. M. (2000). The pragmatics of paediatric language intervention: Issues and analysis. In N. Müller (Ed.), *Pragmatics in speech and language pathology: Studies in clinical applications* (pp. 139–163). Amsterdam: John Benjamins Publishing.

Chapman, R. S., Hesketh, L. J., & Kistler, D. J. (2002). Predicting longitudinal change in language production and comprehension in individuals with Down syndrome: Hierarchical linear modeling. *Journal of Speech, Language, and Hearing Research*, *45*, 902–915.

Ellis, E. M., & Thal, D. J. (2008). Early language delay and risk for language impairment. *Perspectives on Language Learning and Education*, *15*, 93–100.

Ellis-Weismer, S. (2007). Typical talkers, late talkers, and children with specific language impairment: A language endowment spectrum? In S. Ellis Weismer (Ed.), *Language disorders from a developmental perspective: Essays in honor of Robin S. Chapman* (pp. 83–101). Mahwah, NJ: Lawrence Erlbaum Associates.

Gillum, H., & Camarata, S. (2004). Importance of treatment efficacy research on language comprehension in MR/DD research. *Mental Retardation and Developmental Disabilities Research Reviews*, *10*, 201–207.

Gillum, H., Camarata, S., Nelson, K. E., & Camarata, M. (2003). Pre-intervention imitation skills as a predictor of treatment effects in children with specific language impairment. *Journal of Positive Behavior Intervention*, *5*, 171–178.

Haines, J., & Camarata, S. (2004). Examination of candidate genes in language disorder: A model of genetic association in treatment studies. *Mental Retardation and Developmental Disabilities Research Reviews*, *10*, 208–217.

Johnson, C. P., & Myers, S. M. (2007). Identification and evaluation of children with autism spectrum disorders. *Pediatrics*, *120*, 1183–1215.

Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child*, *2*, 217–250.

Kanner, L. (1965). Infantile autism and the schizophrenias. *Behavioral Science*, *10*, 413.

Kjellmer, L., Hedvall, Å., Fernell, E., Gillberg, C., & Norrelgen, F. (2012). Language comprehension in preschoolers with autism spectrum disorders without intellectual disability: Use of the Reynell Developmental language scales. *Research in Autism Spectrum Disorders*, *6*, 1119–1125.

Koegel, R., & Koegel, L. (2006). *Pivotal response treatments for autism: Communication, social, and academic development*. Baltimore, MD: Paul H. Brookes.

Koegel, R., Koegel, L., & Camarata, S. (2010). Definitions of empirically supported treatment. *Journal of Autism and Developmental Disorders*, *40*, 516–517.

Lahey, M. (1988). *Language disorders and language development*. New York: MacMillan Publishing Company.

Lord, C., & Jones, R. M. (2012). Annual research review: Rethinking the classification of autism spectrum disorders. *Journal of Child Psychology and Psychiatry*, *53*, 490–509.

Lord, C., & Risi, S. (2000). Diagnosis of autism spectrum disorders in young children. In C. Lord, & S. Risi (Eds.), *Autism spectrum disorders: A transactional developmental perspective* (pp. 11–30). Baltimore, MD: Paul H. Brookes.

Lord, C., Risi, S., Lambrecht, L., Cook, E. H., Leventhal, B. L., DiLavore, P. C., et al. (2000). The Autism Diagnostic Observation Schedule - Generic: A standard measure of social and communication deficits associated with the spectrum of autism. *Journal of Autism and Developmental Disorders*, *30*, 205–223.

Lord, C., Rutter, M., DiLavore, P., Risi, S., Gotham, K., & Bishop, S. (2012). *Autism Diagnostic Observation Schedule - second edition (ADOS-2)*. Los Angeles, CA: Western Psychological Corporation.

Lovaas, O. I. (1971). Certain comparisons between psychodynamic and behavioristic approaches to treatment. *Psychotherapy: Theory, Research and Practice*, *8*, 175–178.

Lovaas, O. I. (1987). Behavioral treatment and normal educational and intellectual functioning in young autistic children. *Journal of Consulting and Clinical Psychology*, *55*, 3–9.

Lovaas, O. I., Schreibman, L., & Koegel, R. L. (1974). A behavior modification approach to the treatment of autistic children. *Journal of Autism and Childhood Schizophrenia*, *4*, 111–129.

Marcus, L. M., Garfinkle, A., & Wolery, M. (2001). Issues in early diagnosis and intervention with young children with autism. In L. M. Marcus, A. Garfinkle, & M. Wolery (Eds.), *The research basis for autism intervention* (pp. 171–185). New York: Kluwer Academic/Plenum.

Moerk, E. (1992). *A first language taught and learned*. Baltimore, MD: Paul H. Brookes.

Rescorla, L. (2002). Language and reading outcomes to age 9 in late-talking toddlers. *Journal of Speech, Language, and Hearing Research*, *45*, 360–371.

Rescorla, L., & Dale, P., (Eds.). (2012). *Late talkers: Language development, interventions and outcomes*. Baltimore, MD: Paul H. Brookes.

Rondeau, E., Klein, L. S., Masse, A., Bodeau, N., Cohen, D., & Guilé, J. (2011). Is pervasive developmental disorder not otherwise specified less stable than autistic disorder? A meta-analysis. *Journal of Autism and Developmental Disorders*, *41*, 1267–1276.

Smith, A., & Camarata, S. (1999). Using teacher-implemented instruction to increase language intelligibility of children with autism. *Journal of Positive Behavior Interventions*, *1*, 141–151.

Smith, V., Mirenda, P., & Zaidman-Zait, A. (2007). Predictors of expressive vocabulary growth in children with autism. *Journal of Speech, Language, and Hearing Research*, *50*, 149–160.

Snyder-McLean, L., & McLean, J. E. (1987). Effectiveness of early intervention for children with language and communication disorders. In L. Snyder-McLean, & J. McLean, (Eds.), *The effectiveness of early intervention for at-risk and handicapped children* (pp. 213–274). San Diego, CA: Academic Press.

Stone, W. L., & Yoder, P. J. (2001). Predicting spoken language level in children with autism spectrum disorders. *Autism*, *5*, 341–361.

Swedo, S. E., Baird, G., Cook, E. H., Happé, F. G., Harris, J. C., Kaufmann, W. E., et al. (2012). Commentary from the DSM-5 workgroup on neurodevelopmental disorders. *Journal of the American Academy of Child and Adolescent Psychiatry*, *51*, 347–349.

Turner, L. M., & Stone, W. L. (2007). Variability in outcome for children with an ASD diagnosis at age 2. *Journal of Child Psychology and Psychiatry*, *48*, 793–802.

US Centers for Disease Control. (2012). *Autism spectrum disorders: Data and statistics*. Atlanta: Centers for Disease Control and Prevention. http://www.cdc.gov/ncbddd/autism/data.html. Accessed 13 November 2013.

Volkmar, F. R., Cohen, D. J., & Paul, R. (1986). An evaluation of DSM-III criteria for infantile autism. *Journal of the American Academy of Child Psychiatry*, *25*, 190–197.

Warren, Z., McPheeters, M. L., Sathe, N., Foss-Feig, J., Glasser, A., & Veenstra-VanderWeele, J. (2011). A systematic review of early intensive intervention for autism spectrum disorders. *Pediatrics*, *127*, 1303–e1311.

Wetherby, A. M., Woods, J., Allen, L., Cleary, J., Dickinson, H., & Lord, C. (2004). Early indicators of autism spectrum disorders in the second year of life. *Journal of Autism and Developmental Disorders*, *34*, 473–493.

Whitehurst, G., Fischel, J., Arnold, D., & Lonigan, C. (1992). Evaluating outcomes with children with expressive language delay. In S. Warren, & J. E. Reichle (Eds.), *Causes and effects in communication and language intervention. Communication and language intervention series*, *Vol. 1.* (pp. 277–313). Baltimore, MD: Paul H. Brookes.

Whitehurst, G. J., Fischel, J. E., Lonigan, C. J., Valdez-Menchaca, M. C., Arnold, & D. S., Smith, M. (1991). Treatment of early expressive language delay: If, when, and how. *Topics in Language Disorders*, *11*, 55–68.

Yoder, P. J., & Warren, S. F. (1993). Can developmentally delayed children's language development be enhanced through prelinguistic intervention? In A. P. Kaiser, & D. B. Gray (Eds.), *Enhancing children's communication* (pp. 35–61). Baltimore, MD: Paul H. Brookes.

Zwaigenbaum, L., Thurm, A., Stone, W., Baranek, G., Bryson, S., Iverson, J., et al. (2007). Studying the emergence of autism spectrum disorders in high-risk infants: Methodological and practical issues. *Journal of Autism and Developmental Disorders*, *37*, 466–480.