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Minimally Verbal School-Aged Children with Autism Spectrum Disorder: The Neglected End of the Spectrum

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Abstract

It is currently estimated that about 30% of children with autism spectrum disorder remain minimally verbal, even after receiving years of interventions and a range of educational opportunities. Very little is known about the individuals at this end of the autism spectrum; in part because this is a highly variable population with no single set of defining characteristics or patterns of skills or deficits, and in part because it is extremely challenging to provide reliable or valid assessments of their developmental functioning. In this paper we summarize current knowledge based on research including minimally verbal children. We review promising new novel methods for assessing the verbal and nonverbal abilities of minimally verbal school-aged children, including eye-tracking and brain imaging methods that do not require overt responses. We then review what is known about interventions that may be effective in improving language and communication skills, including discussion of both non-augmentative and augmentative methods. In the final section of the paper we discuss the gaps in the literature and needs for future research.

Research in the field of autism spectrum disorder (ASD) has flourished over the past two decades. However, the vast majority of studies have focused mostly on either young toddlers and preschoolers or older higher functioning, verbal children, primarily because they are easier to evaluate using standard assessment tools and they are more compliant during behavioral or neuroimaging experimental investigations. Recently, the Interagency Autism Coordinating Committee highlighted the dearth of knowledge about nonverbal children with ASD (IACC 2011 Strategic Plan: <http://iacc.hhs.gov/strategic-plan/2011/index.shtml>). As awareness about this issue grew in recent years, Autism Speaks held a series of meetings in 2009 on “Characterizing cognition in nonverbal individuals with autism” and the National Institutes of Health convened a workshop that was held in April 2010 to identify what is currently known, what are the gaps in our knowledge, and what are the research opportunities that could address these gaps. In this paper, which grew out of the NIH workshop, we summarize current research on minimally verbal school aged children with ASD, focusing on three main questions: (1) Who are these children? (2) What novel technologies could be used to assess their receptive language and other cognitive skills? (3) Which interventions may be effective in improving their language and communicative skills?

(1) Minimally verbal children with ASD

It is not known how many children with ASD remain with little expressive spoken language abilities by the time they reach school age. Older statistics suggest that over half of all children with autism failed to acquire spoken language (National Research Council, 2001), however more recent studies suggest that this figure is now lower, at around 30%, in part because of the broadening of diagnostic criteria, in part because more verbal children are now identified as having autism, and in part because of earlier diagnoses as well as greater access to more effective early interventions that significantly improve spoken language and communication skills in younger preschoolers with ASD, thus potentially preventing them from remaining nonverbal at later ages (Tager-Flusberg, Lord & Paul, 2005).

We do not understand why, despite access to interventions, some children fail to make progress in acquiring language, but what is clear, is that this group of children is extremely variable and therefore no single explanation will account for all minimally verbal children. While some children slowly acquire language during the preschool years, estimates are that about two-thirds to three-fourths of all children will have some words, and that at least half will be able to use phrase speech by the time they enter primary school (Anderson et al, 2007; Magiati, Moss, Charman & Howlin, 2011; Turner, Stone, Pozdol, & Coonrod, 2006). Thus, while most preschool aged children with ASD are *preverbal*---they will eventually learn to use spoken language---about 25–30% will be *nonverbal, or only minimally verbal* by the time they enter Kindergarten (Anderson et al, 2007). Given the goal that “useful speech by age 5” consistently predicts better social and adaptive functioning later in life, the Interagency Autism Coordinating Committee set a long term goal that 90% of children with autism would acquire useful speech by age 5 (Department of Health and Human Services, 2004). Thus, most efforts at increasing spoken language have been focused on children younger than 5 years of age.

The distinction between preverbal and nonverbal may be significant but at this point is difficult to determine. One issue is the lack of clear definitions of preverbal and nonverbal. Take for example definitions of ‘nonverbal’ preschool aged children for different intervention studies. Ronski et al (2010) defined nonverbal toddlers as those whose Mullen expressive language scores were below 12 months and had fewer than 10 intelligible spoken words. Yoder and Stone (2006) defined their preschool participants as non- or low verbal based on fewer than 20 different words used over three separate language samples. Koegel et al (2009) included preschool aged children with no functional words and no object-label correspondence and Kasari et al (2008) defined as low verbal 3–4 year old children who had fewer than 5 spoken words based on the combination of observational and standardized assessments. Given that toddlers and preschoolers are chronologically young, the extent to which they may be just *preverbal* (they are delayed in their language now but will use spoken language in the near future) versus *nonverbal* (they do not use spoken language now and will continue to not use spoken language in the near to far future) is unclear.

The literature on minimally verbal children with ASD is quite sparse and there is little consistency in the definition of this group. In some cases these individuals lack all spoken language: their vocalizations only include atypical non-speech sounds and some vowel approximations. In other cases their expressive language is extremely limited with just a few words or fixed phrases (e.g., *Want X*) used infrequently and only in limited contexts. Other children included in this group may have some spoken language, but they are primarily echolalic or use stereotyped or scripted language in ways that appear non-communicative. Some children who do not speak are able to use alternative means for communicating to varying degrees of proficiency, including sign language, PECS (Picture Exchange Communication System), VOCA (Voice Output Communication Aids) or even written

language, suggesting that these children may vary in their preference for relying on visual or auditory modalities for communicating with others.

One common assumption is that all children who fail to acquire spoken language have low nonverbal IQ scores. However, a recent large-scale study of preschoolers found that while some minimally verbal children have low nonverbal IQ, others do not (Munson et al., 2008). Similarly, while some minimally verbal school aged children have low receptive and expressive language skills, other children with minimal expressive language scores have good receptive language abilities, which are correlated with their nonverbal abilities (Rapin et al., 2009). These examples of heterogeneity in cognitive and linguistic skills among minimally verbal children with ASD provide support for the view that no single underlying mechanism will explain why these children do not learn to speak.

Few studies have investigated factors that predict who remains nonverbal beyond the early preschool years. Retrospective parent report data suggest that infants who are later diagnosed with autism and fail to acquire spoken language show very significant delays in developing oral motor skills during the first year of life (Gernsbacher et al., 2008). The majority of studies focusing on predictors of spoken language have investigated toddlers and preschoolers. The ability to imitate sounds and simple movements is one key predictor of expressive language (e.g., Luyster et al., 2008; Stone & Yoder, 2001; Thurm et al., 2007). Response to joint attention predicts receptive language (Anderson et al., 2007; Luyster et al., 2008; Sigman & McGovern, 2005), and nonverbal cognitive abilities predict both expressive and receptive language (Luyster et al., 2008; Thurm et al., 2007). In some cases, the almost complete absence of any social motivation may be associated with no spoken language. As these nonverbal children get older, about 25% begin to show more significant adverse consequences of having no means for communicating, with increased social withdrawal on the social lethargy subscale of the Aberrant Behavior Checklist (Lord, 2010). These findings underscore the urgent need to begin investigating what the underlying mechanisms are that prevent some children with ASD in acquiring spoken language, and what the possibilities are for interventions implemented at older ages.

In the literature on language acquisition, the benchmark of acquiring some spoken language by the age of five is considered highly significant; thereafter it is unlikely that a child would acquire significant linguistic skills (e.g., Tager-Flusberg et al., 2005). Nevertheless, it is now clear that some children with ASD do begin speaking after this point, as highlighted in a review by Pickett and her colleagues (Pickett et al., 2009). They surveyed the literature from 1951–2006 and identified 167 cases of children who started speaking after age 5. The majority began talking between the ages of 5 and 7, and some at even older ages, up to age 13. These children had some conceptual and semantic abilities that allowed them to map sounds to meaning, and were more likely to have nonverbal IQ scores over 50. All the successful children at these older ages seemed to be motivated to communicate orally, and had received intensive, often behaviorally based, training in the formation of sounds and words. While the majority of these children increased their single word utterances, only about one-third moved into phrase speech.

Gordon (2010) conducted a case study of one nonverbal boy with ASD who began a highly intensive home-based intervention with experienced clinicians at the age of 12. He progressed from having just a single word at age 12 to acquiring a relatively large single word vocabulary and multi-word utterances through the adolescent years. At age 22, this young man continued to show progress in spoken language. The intervention program was flexibly implemented and multi-faceted. Initially the focus was on speech-articulation skills, borrowing techniques from the adult aphasia literature. Over time the program was changed frequently, addressing the specific individual needs of the young man. The significance of

this case study is in demonstrating what might be possible during the adolescent years, though some caution is warranted as it is unlikely that the same progress could be replicated across the majority of nonverbal individuals with ASD.

(2) Novel technologies for assessing receptive language and cognition

Most of the published studies on older minimally verbal children with ASD fail to provide much information about the participants other than age and expressive language level (cf. Pickett et al., 2009). One significant barrier is the dearth of valid, reliable and appropriate means for direct assessments of this population. Instead, most studies that do report on some characteristics of their sample rely on parent report measures (questionnaires or interviews) rather than clinical testing. Standard methods for assessing even foundational cognitive or receptive language skills depend on a range of behaviors that may not part of the repertoire of the minimally verbal child. These include the ability to develop rapport with the examiner, the motivation to comply with task demands, capacity to understand the pragmatics of the testing situation, attention or interest in the testing materials, interference from challenging behaviors, anxiety or frustration, and basic responses such as pointing skills (Tager-Flusberg, 1999). For all these reasons it is often not possible to conduct direct assessments using currently available standardized tests (but see the companion paper on current options for assessing this population; Kasari, Brady, Lord and Tager-Flusberg).

With the advent of newer technologies that have been applied successfully to other nonverbal populations such as preverbal infants, novel methods for assessing minimally verbal children with ASD are now potentially available. These include the use of eye-tracking measures (cf. Aslin, 2007; Aslin & McMurray, 2004), neurophysiological measures including event-related potentials (ERP) or electroencephalography (EEG; Choudhury & Benasich, 2011; Connolly et al., 2006), and magneto-encephalography (MEG; Imada et al., 2006), which are more feasible to implement than other brain imaging methods. At this point, most of these methods, which do not require the child to understand or follow even minimal instructions and do not require an overt motor response, are in the development phase for studies on minimally verbal children with ASD, with several research groups evaluating their reliability and validity. Thus far, work has begun to evaluate these implicit methods as ways of assessing minimally verbal children's receptive language or auditory processing skills. While they seem to hold considerable promise as useful measures, not every child can comply with even the minimal demands of these methods, all of which require some degree of motion control.

a) Eye-tracking

Looking behavior has been used extensively in studies of infants' cognitive and linguistic knowledge (Aslin, 2007; Aslin & McMurray, 2004), as well as in research on visual preferences in children and adults with ASD (e.g., Boraston & Blakemore, 2007; Chawarska et al., 2010; Klin et al., 2002; Pelphrey et al., 2002). With the advent of newer models of eye-tracking systems, it is now possible to collect accurate recordings of looking time without requiring any apparatus to be mounted on the child's head or other invasive procedures. Instead, these systems (e.g., Tobii eye-tracking systems; <http://www.tobii.com/>) rely on infra-red sensors that are embedded in the computer monitor to capture corneal reflection, which is the basis for detecting a person's point of gaze. Calibration is fast and easy and does not require the participant to follow verbal instructions; it is achieved by placing attention-grabbing icons on the computer screen that can be followed automatically. The method does require that the content of the computer screen before and during any task administration is sufficiently engaging for children to maintain attention without them moving out of the range of the embedded sensors.

To assess receptive language, the basic assumption of eye-tracking measures is that when a child hears a word, phrase or sentence, he or she will locate and then look longer at an image (picture, cartoon, or video) that matches the target language than at an unrelated image placed next to the matching image. If a child looks reliably longer at the matching image (ensuring through counterbalanced presentation that the child does not show a significant bias to looking mostly at one side of the screen), then it is assumed that the child understands the linguistic stimulus. Current studies are underway by Tager-Flusberg and her colleagues (e.g., Edelson et al., 2008) and others to establish that for minimally verbal children with ASD, eye-tracking measures provide a valid assessment of receptive language. Thus far, lexical comprehension has been tested using parent information for which words a child understands. Preliminary data indicate that, like verbal children with autism and typically developing children, minimally verbal children with autism look longer at matching pictures for words that they know, but fail to do so for words they do not understand (Edelson, et al., 2008).

b) Neurophysiology

For over 30 years, ERPs have been used to index different aspects of language comprehension as well as other aspects of cognitive functioning (Kutas & Federmeier, 2011). While most research has been conducted with typical adults, ERPs have been applied to other populations including preverbal infants (e.g. Friedrich & Friederici, 2010), children with language and other neurodevelopmental disorders (Bishop, 2007), or adults who have suffered strokes or other neurodegenerative disorders (e.g., Byrne et al., 1999; Kotz & Friederici, 2003). Studies of ERPs in verbal children with ASD have investigated language and other cognitive processes, such as face recognition (for review, see Jeste & Nelson, 2009). ERPs reflect the activity of populations of neurons (primarily from pyramidal cells) time-locked to specific stimulus events. Because this activity is collected at the skull surface, brain activity must be averaged over a relatively large number of trials in order to achieve a reasonable signal-to-noise ratio. ERPs provide sensitive measures of temporal (but not spatial) aspects of stimulus processing, and specific components of an ERP provide measures of distinct processing stages, for example perceptual, semantic or grammatical aspects of language.

Electrophysiological responses are collected from electrodes attached to the head. Ideally, placing the electrodes, which can vary in number depending on the system or specific research paradigm, would be accomplished in a minimal amount of time to make this method feasible and more easily tolerated, especially by minimally verbal children with ASD. The use of an electrode cap or net significantly reduces the time needed for attaching electrodes. No matter how quickly electrodes are placed, with or without a cap or net, they may be quite uncomfortable for some children. It is therefore important to develop a training protocol that might take several sessions that would gradually introduce and desensitize the child to the electrode system. While some movement is possible during the collection of electrophysiological measures since data can be corrected for a certain degree of motion, data collection during stimulus presentation should be accomplished with minimal movements, especially of the head and eyes since trials on which there is movement or eyeblinks must be discarded.

As noted, different components of the ERP reflect different aspects of linguistic processing (for review, see Friederici, 2005). Lexical meaning can be assessed using the N400 (a negative wave, between about 250 and 600 msec, usually peaking at about 400 msec after stimulus onset, collected from electrodes placed over central/parietal regions). The amplitude of the N400 will be reduced, if prior to hearing the word, a semantically-related prime (e.g., picture or word) is presented. This difference in the N400 amplitude can be taken as an index of lexical comprehension. For morpho-syntactic processes there are both

early (left anterior negativity or LAN, occurring 200–400 msec post stimulus) and late (P600 positive wave form peaking at around 600 msec) components of the ERP that are elicited if a person detects ungrammatical speech, for example incorrect plural/tense marker (e.g., **The girls dances*). Meaningful connected speech is distinguished from non-meaningful connected speech by a slow positive component primarily over temporal lobe sites. To collect these data, children just need to look at the computer screen (if visual stimuli are used) and listen passively to the linguistic stimuli; no overt response is needed.

These paradigms are all currently being implemented by Benasich and her colleagues in studies of both younger and older minimally verbal children with ASD (Benasich et al., 2011). With training and preparation, the majority of participants were able to provide useful data (see also Connolly et al., 2011). In preliminary findings, Benasich et al. (2011) reported that some minimally verbal children showed the expected ERP responses, while others did not. These findings suggest that neurophysiological measures can distinguish between children who do understand speech from those who fail to, though it should be noted that not all participants with ASD were able to provide sufficient artifact-free data for analysis.

c) Magnetoencephalography

MEG is another brain imaging tool that provides sensitive information about brain processing of stimuli by measuring electromagnetic brain signals from the cortex (for review, see Hari et al., 2010). MEG records dipolar magnetic fields produced by populations of neurons firing synchronously. Like ERPs, MEG offers sensitive measures of temporal aspects of neural processing; when structural images of the brain are also available then MEG can also provide excellent spatial resolution. A MEG system is, however, far more expensive than EEG, not portable, and is generally not available outside major research centers. Most MEG machines are built for adult use, and may require adaptation for use with younger children (Imada et al., 2006).

A few studies have used MEG to investigate auditory (e.g., Gage et al., 2003; Roberts et al., 2010) or visual processing (e.g., Bailey et al., 2005) in ASD, though its primary use has been to investigate impairments in neural mechanisms in adolescents or adults ASD (and other disorders) rather than as a non-invasive tool for assessing cognitive or linguistic function. Nevertheless, because passive paradigms can be implemented with MEG, it has some potential to be used for these purposes, as is the case with ERPs (Gage et al., 2011). For MEG to be used effectively with minimally verbal children, training for motion control and tolerance of the machine are needed, though unlike functional MRI, there is no noise associated with the machine, and the participant can be seated upright in greater comfort without being encased in a tube.

d) Evaluation of novel methods of assessment

Given the significant challenges inherent in the assessment of cognitive, linguistic, and behavioral skills in the minimally verbal child with ASD, there is a need for new methods that involve exclusively passive paradigms, no overt responding, or even the ability to understand instructions. Several tools are now available that rely on eye movements or neural measures of response to stimuli that seem promising, at least for some children. Nevertheless, we are still a long way from having psychometrically sound measures that take advantage of these new tools. For each of the technologies reviewed here there are advantages and disadvantages. For all of them, children must receive training to tolerate the testing environment, and minimize movement. Tasks must be designed carefully to ensure that they capture specific perceptual, cognitive or linguistic processes, engage the attention of the child and maintain the child's interest. Methods must provide meaningful measures at the individual child level; this is currently more easily achieved for eye-tracking and MEG

than for ERP, which relies mostly on group comparisons, averaging across participants. For all these technologies, there is a trade-off between having a sufficiently large number of valid trials (yielding usable, artifact-free data) and the length of a testing session. Eye-tracking can be used to assess a wide range of linguistic processes, but some children will fail to look at the computer screen or may show a bias to attend only to one side. For paradigms that rely on subtle timing of eye movements as a measure of linguistic knowledge, there may be some concern that oculomotor abnormalities that have been identified in some children with ASD will affect interpretation of data (e.g., Takarae et al., 2007). While MEG provides the richest data on temporal and spatial processing from cortical systems and is well suited for single subject data collection, it is expensive and currently less well developed for capturing specific stages of linguistic processing than ERPs. We return to discussing the needs for further development of these novel methodologies in the final section of this paper.

(3) Interventions for the minimally verbal child with ASD

Most children beyond preschool age are not completely nonverbal; they often have some words or phrases that may or may not be used communicatively. Our understanding of how well these children respond to interventions depends on who is included in intervention studies, and how we define pre-treatment characteristics. Currently we have limited information on children who may have the most difficulty with spoken language. Intervention studies often exclude ‘low functioning’ potentially nonverbal children. For example, studies of comprehensive early interventions (e.g., Dawson et al, 2009; Lovaas, 1987) have excluded children with developmental quotients (or IQ scores) below 35 or developmental ages below 12 months. Intervention studies of older, school-aged children often focus on verbal higher functioning children (e.g., improving pragmatic skills), also excluding children who are minimally verbal. As a result, we know little about the heterogeneity in language learning of a broad spectrum of children, particularly during transition periods, from preschool to school age, childhood to adolescence, and adolescence to adulthood.

a) Interventions targeting spoken language outcomes in school-aged children

For the school-aged minimally verbal child the focus is often on functional communication outcomes. Several studies have used naturalistic behavioral methods, such as incidental teaching to increase spoken language and functional communication outcomes. One example in this line of work found that, compared to discrete trial training, creating more opportunities for incidental teaching via parents was associated with greater success in increasing verbal responses in three nonverbal boys between 6 and 9 years of age (Charlop-Christy et al., 2002). The boys in this study learned to say and generalize set phrases, such as, “*Good morning*” but there is no evidence they could spontaneously produce non-targeted words or phrases. Other studies have used functional communication training to decrease problem behaviors and increase communication (Mancil et al, 2009). In nearly all cases, interventions target the training of words or phrases that are often set phrases or words used to request, “*I want...*”. Thus, in these studies changes in communication were limited to the requesting function and increased responding, not spontaneous initiations.

b) Interventions using augmentative and alternative communication (AAC)

There is currently considerable emphasis on the use of augmentative and alternative modes of communication for school-aged children who are minimally verbal. AAC systems encompass any non-speech means for expressive and receptive communication including sign language, visual systems, and speech generating devices (e.g., VOCA). The growing availability of the iPad and other tablets, for example, has generated a great deal of

enthusiasm for their potential to help minimally verbal individuals communicate. However, more commonly applied AAC systems rely on ‘low-tech’ solutions such as the Picture Exchange Communication System (PECS). PECS is an augmentative communication system in which children exchange pictures in order to communicate with others and is widely applied in classrooms of children with ASD.

In the only randomized clinical trial (RCT) including children over age five, the effectiveness of PECS for 84 children was assessed (Howlin et al, 2007). In this study, randomization was at the unit of the classroom, including all children in the class between 4 and 10 years of age who were reported to have limited language based on the Autism Diagnostic Observation Schedule diagnostic assessment. The main findings were that the children in classrooms whose teachers, parents and staff had received PECS training initiated communication at a higher rate using PECS, but there was no main effect on the use of spoken language. Indeed, there is little evidence that PECS increases the production of spoken language (Preston & Carter, 2009). One exception is a study that used a single-subject multiple baseline design in which 2 of the 3 children were over age 5 (Charlop-Christy et al, 2002). Both the older children increased their use of requesting with the picture cues and also increased their spontaneous production of spoken requests. Thus, PECS does not appear to *inhibit* the development of spoken language, but there is only limited evidence that the use of PECS *facilitates* increased spoken language (Schlosser & Wendt, 2008). One further limitation in all these studies is that when language is targeted or is reported to improve, the language function is nearly always limited to requesting.

Despite single subject studies of successful use of AAC speech generating systems for increasing communication and spoken language in some individuals (Schlosser & Wendt, 2008; Sigafoos et al, 2004), researchers report very limited use of AAC devices either in classrooms or at home (Brady, 2010). One issue may be the lack of specific teaching strategies that can be utilized by teachers and parents when they have access to an AAC system. Unlike PECS, which has a specific sequence of teaching children how to use the system, speech generating devices do not have a similar teaching package for teachers. Given new technologies providing speech output (e.g., tablets with the proloquo2go application), development of interventions using these devices is urgently needed.

c) Application of preschool interventions to school-aged children

Since there has been little in the way of developing specific interventions for older, minimally verbal children, an alternative approach is to apply or adapt evidence-based interventions that have been designed and evaluated on younger children with ASD. The younger developmental age of preschoolers with autism may be commensurate with the skill levels of older minimally verbal children, such that adapting effective early interventions for older children may prove beneficial. One issue to consider is whether these same interventions that have been evaluated in young, essentially *preverbal* children will work equally well with older children who are minimally verbal.

Several studies have found that preschool-aged children with limited spoken language skills become verbal from exposure to augmentative communication approaches including PECS, sign language or speech generating devices (e.g., Ronski et al, 2010; Sulzer-Azaroff et al, 2009). These outcomes with younger children appear stronger than those obtained from older minimally verbal children. For example, one RCT with preverbal toddlers of mixed etiologies reported increased vocabularies when these children were using speech-generating devices (Ronski et al, 2010). Toddlers who received interventions of speech input or output using a speech-generating device had larger spoken and augmented vocabularies compared to toddlers who received only standard speech therapy. Thus, these data suggest significant benefits of speech generating devices in increasing communication (spoken and augmented)

at least over a short period of time in preschool-aged children. A reasonable question is whether intervention strategies in these studies are different from those applied to older children and if so, are they applicable to older children so that they might strengthen treatment outcomes in the minimally verbal older child?

Other studies of preverbal preschoolers have focused more directly on spoken language. Using naturalistic behavioral approaches, specifically, pivotal response training (PRT), Koegel et al, (2009) found that 3 and 4 year old children with ASD acquired spoken words if they were cued to attend (e.g., using a ‘high five’ gesture) prior to being prompted to say a word. Thus, recruiting the children’s attention and then delivering the word prompt yielded significant improvement in producing words. These young children had failed to respond to traditional behavioral treatments, and had been considered “non-responders” to other interventions. In another study using a combination of naturalistic, developmental and behavioral strategies, 3 and 4 year old children with autism were randomized to one of three conditions: joint attention intervention, symbolic play intervention or control ABA only (Kasari et al, 2006). Children in all three conditions were simultaneously receiving 30 hours of ABA center-based preschool services. The two experimental interventions (joint attention and symbolic play) not only yielded significant improvements in joint attention and play skills, they also predicted better expressive, spoken language one year later (Kasari et al, 2008). In a child by treatment interaction, the study found that children who began treatment with fewer than 5 spoken words in their repertoire made the most gains in spoken language if they received the joint attention intervention. Taken together, these studies suggest that specific strategies (e.g., cueing for attention), content (e.g., joint attention) and mode of communication (e.g., augmentative) may all affect the production of spoken communication in preschool aged children who begin treatment with limited or no spoken words; it remains to be seen if the same strategies could be effective in promoting speech in older minimally verbal children.

d) Factors associated with communication outcomes

A number of pretreatment child characteristics may help in predicting language outcomes and point to potential treatment targets. Several studies have found that joint attention skills (both responding and initiating) predict later language acquisition. For example, Sigman and Ruskin (1999) reported that responding to joint attention predicted spoken language up to 10 years later in a sample of children with autism who were originally assessed during the preschool years. Another important predictor of language outcome might be the ability to use even a single word. In an ongoing RCT with preschoolers comparing PECS (visual approach) with PRT (verbal approach) one of the predictors of better outcomes in both conditions was if a child had one or more words prior to treatment (Schreibman, 2010). In another study, even a minimal amount of language predicted spoken language gains in children who began PECS treatment between 4 and 10 years of age (Gordon et al, 2011).

Other factors could also play a significant role in language outcomes. In examining the prediction to language over the course of two years for children who began intervention when they were about 4 years-old, Bopp et al. (2009) found two factors were significant: inattentiveness and social motivation. Children who scored high on measures of inattentiveness and demonstrated limited social motivation made far less progress in acquiring language. These findings were found independent of the children’s initial autism severity scores and nonverbal IQ. Interestingly, behaviors one would expect to interfere with language development, such as acting out or repetitive behaviors were not associated with language outcomes. Sherer and Schreibman (2005) found that the best responders to PRT interventions were those children who were more engaged with objects, had some verbal behavior (echolalia), more social approach and less avoidance behavior. Thus, directly

targeting problems in social motivation, attentiveness and object play skills may be important targets for children who do not make significant progress in developing language.

e) Summary and evaluation of interventions research

Current intervention research for the minimally verbal older child with ASD has mostly focused on behavioral and naturalistic interventions aimed at encouraging spoken language with or without the assistance of AAC systems. To date, there is scant evidence for effective language interventions for these children. Most success has been found in helping children increase requesting initiations and responses, primarily in studies using visually based augmentative systems such as PECS, but there are no data on improvements in other communicative functions, such as commenting. There is also limited spoken language change as a result of AAC interventions with older children.

One issue to tackle is the heterogeneity in the definitions of ‘nonverbal’ and in assessments of child characteristics prior to intervention. Knowing more about the characteristics of the children enrolled in intervention studies across a wide range of behaviors will allow us to understand better child response to treatments, thus giving us the potential for creating individualized treatments for optimal outcome. The data from studies of younger children suggesting that attentiveness, social motivation, object play skills, and even a limited vocal repertoire indicate that all these factors predict better communication outcomes. These are components of interventions that may need further development for older minimally verbal children with ASD.

(4) Future research

This overview of current research on minimally verbal children with ASD has highlighted how little we know about them. There is a clear need to define a set of research agendas that can address the many gaps in our knowledge. So few publications focus specifically on the older minimally verbal child; in our view, children below the age of 5 who are not speaking should be considered *preverbal*, as they still have the potential to acquire spoken language skills given appropriate interventions. We do not know whether children over the age of 5, are similar to younger children, but with more profound impairments in autism and related symptoms, or whether there are qualitative differences in these minimally verbal children at the genetic, neurobiological or behavioral levels (e.g., Flax et al., 2010). Research studies addressing the full scope of issues raised by this end of the ASD spectrum are clearly needed. We outline here a number of questions that need to be answered if we are to begin to meet the urgent needs of these individuals.

- a. In what ways are minimally verbal children with ASD different from minimally verbal children with intellectual disability (without ASD)? While some of the children with ASD who do not learn to speak also have severe levels of intellectual disability, it seems that some do not; for any given level of intellectual disability, the child with ASD is more likely to remain nonverbal. Studies that compare these populations may reveal the specific characterization and significance of social motivation or other factors unique to ASD for acquiring spoken language skills.
- b. Nothing is known about the mechanisms that explain why some children with ASD do not learn to speak. There are hints in the literature that oral-motor skills (e.g., Gernsbacher et al., 2008), auditory processing skills (e.g., Gage et al., 2011) or even genetic factors (e.g., Flax et al., 2010) differentiate this group, but there are still no studies that specifically address this core issue. We also know little about how children with ASD may make use of alternate routes to spoken language. At the neurobiological level, some children may acquire spoken language relying more on the right hemisphere than on the more typical left hemisphere for processing

language (e.g., Knaus et al., 2010; Redcay & Courchesne, 2008); others may acquire language through reading rather than in the spoken form (e.g., Bryson et al., 1994). As we make progress in identifying the underlying mechanisms or alternative pathways, the potential to discover predictors in infancy or early childhood will be possible, leading eventually to preventive treatments and interventions that may be implemented during this optimal period of development.

- c. Studies targeting minimally verbal children with ASD, including those addressing core mechanisms or potential treatments, should provide comprehensive evaluations of each child across multiple behavioral domains in a range of social contexts. Although such assessments may be difficult to accomplish, investigators should attempt to examine behaviors that are known precursors of language (including object play, joint attention skills, intentional behaviors, imitation, vocalization and nonverbal cognitive abilities; see companion paper, this issue, by Kasari et al.). New technologies for assessing cognitive and linguistic abilities should be further developed with particular attention paid to developing psychometrically valid measures based on implicit measures derived from eye-tracking or neuroimaging methodologies. Adaptations of current standardized tests on tablets or touch screen computers might also be more effective methods for assessing this population by capitalizing on their motivation and engagement with these technologies.
- d. New research paradigms are needed for intervention studies that for example, integrate assessment with intervention. In conducting future research studies, investigators will likely need to use creative research designs such as response to treatment (RTI) or sequential treatment designs. Treatment as usual designs are not as informative if children have already had access to adequate intervention but had made limited spoken language progress. Rather than offering more of the same, researchers need to employ comparative treatment designs to disentangle the active ingredients of intervention and to determine which children benefit most from a specific intervention, or sequence of tailored intervention strategies. There is also the potential for incorporating novel implicit and brain imaging measures to assess changes that may occur prior to observable behavior changes during the course of an intervention, and to assist us in better understanding the mechanisms of treatment change.
- e. Intervention studies with older children have generally relied on case studies or single subject designs, and the participants enrolled in these studies have not been adequately characterized. Randomized controlled trials are rare. A critical consideration is that the comparison to a no-treatment control group, or practice as usual, may not be appropriate with older children already exposed to current practices and not progressing as expected. There is some promise in the application of alternating treatment designs, such as SMART (sequential multiple assignment randomization trial) designs that should be considered in future studies for this population (Collins, Murphy & Strecher, 2007).
- f. Studies are needed that examine the transition from preverbal to verbal as children move into the school years. Particularly around the age of 4 to 5 years, there may be specific developmental changes that place a child on a path to becoming verbal or to remain only minimally verbal. Novel interventions that target this transitional stage may be important to develop with the goal of improving the language outcomes of this group.
- g. New technologies are rapidly being introduced for use as interventions for minimally verbal children. In particular a growing number of applications are being

developed for the iPad and other tablets that are receiving considerable attention in the media and from consumers. But little research has been conducted to evaluate these technologies, and most do not have systematic curricula associated with them that would potentially enhance their efficacy. Studies are needed that would fill the gaps in our knowledge about how these technologies might be used most effectively and what the limits might be for implementing interventions on these platforms for this population. Novel language interventions could also be developed that capitalize on a child's interest and strengths in other areas. For example, music might serve as bridge and facilitate spoken language in some nonverbal children (Wan & Schlaug, 2010).

Conclusions

We have clearly made considerable progress in understanding early predictors of language acquisition and the development of effective interventions that target toddlers and preschoolers with ASD. In contrast, we know almost nothing about minimally verbal older children, children who despite access to these interventions fail to acquire spoken language skills. We hope that this review will stimulate the research community to begin to address some of the pressing needs for this group of individuals who, for too long, have remained at the neglected end of the autism spectrum.

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