

Differential language markers of pathology in Autism, Pervasive Developmental Disorder Not Otherwise Specified and Specific Language Impairment

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ABSTRACT

Background: Language impairment is a common core feature in Pervasive Developmental Disorders (PDD) and Specific Language Impairment (SLI). Many studies tried to define their specific language profiles, some claiming the existence of overlaps between the disorders, and others conceiving them as separate categories. Fewer sought to determine whether and how PDDNOS language profile differs from those of Autistic Disorder (AD) and SLI.

Objective: The aim of our study was to (i) document the language profiles of three developmental disorders (AD, PDD NOS, SLI) by assessing phonology, vocabulary, syntax, pragmatics and prosody and (ii) determine differential language markers of pathology.

Method: Twelve children with AD (mean age 9,75; sd 3,5), 10 with PDDNOS (mean age 9,83 ; sd 2,17), and 13 children with SLI (mean age 9,17 ; sd 3,9) were recruited and matched for age, sex and academic skills. To explore both receptive and expressive skills with used “l’examen du langage oral” [Assessment of Oral Language]. To explore prosody, a prosodic imitation task was created and analyzed through automatic speech processing and compared to 70 typical developing controls matched for age and sex.

Results: We observed a similar delay in phonology and vocabulary in the 3 groups and significant but variable differences between the groups in syntax, pragmatics and prosody. SLI showed correlations between chronological age and raw scores in all language tasks, while AD and PDDNOS did not. Furthermore, in SLI, all raw scores in language tasks were correlated within them. Most of those correlations were also found in PDDNOS, while it was not the case in AD.

Conclusion: Our findings support the hypothesis that language skills and development in AD and SLI rely on different mechanisms, while PDDNOS show an intermediate profile, sharing some characteristics of both AD and SLI. They also suggest that expressive syntax, pragmatic skills and some intonation features could be considered as language differential markers of pathology.

Keywords: Autism, Specific Language Impairment, Pervasive Developmental Disorder Not Otherwise Specified, Language, Prosody.

INTRODUCTION

Pervasive Developmental Disorders

Pervasive Developmental Disorder (PDD) is characterized by a triad of severe deficits and pervasive impairments in developmental areas such as reciprocal interactions, communication skills and stereotyped behavior, interests and activities (Wing and Gould, 1979). PDD is mostly seen as a “spectrum disorder”, with several variants and gradients. DSM IV (APA, 2000) encompasses the broad range of individuals with deficits in these three behavioral domains and describes three main disorders: i) Autistic Disorder (AD) with early onset, possible mental retardation, language impairment and symptoms in all areas characterizing PDD; ii) Asperger Syndrome (AS) without language delay or mental retardation; iii) Pervasive Developmental Disorder Not Otherwise Specified (PDDNOS) in which social, communicative and/or stereotypic impairments do not coexist, or in a lesser degree, resulting in that they do not fit other subtypes of PDD. Thus, diagnostic criteria for PDDNOS are quite vague and mostly lead to a diagnosis by default (Volkmar, 2005), while being statistically the largest category (Fombonne, 2003).

Language in PDD

Kanner (1943) and Asperger (1944) described language abnormalities such as mutism, echolalia, pronoun reversal, poor relation to conversational context, lack of drive to communicate, idiosyncrasy, verbosity and aberrant prosody. Language and communication impairment in PDD, especially in AD, led to numerous studies over the last decades, trying to specify profiles. Language in autism, when present, may show varying several subtypes within the spectrum (Kjelgaard & Tager-Flusberg, 2001 ; Rapin & Dunn, 1997; Tager-Flusberg, 2006). Some individuals may have *structural language disturbances*: i) delayed phonology, especially in non-word repetition tasks (Tager-Flusberg, 1981; Bartolucci et al, 1976; Kjelgaard & Tager-Flusberg, 2001 ; Whitehouse et al, 2007) ; ii) poor comprehension skills, sometimes more impaired than the expressive ones (Boucher, 2003 ; Rapin and Allen, 1997 ; Bartak et al, 1977 ; Tager-Flusberg, 1981), iii) immature syntax and prevalence of syntactic errors (Kjelgaard & Tager-Flusberg, 2001). Functional deficits are characterized by: i) a core pragmatics disorder (defined as the ability to use and understand the rules governing language as a communicative tool, including tone of voice, facial expressions, communicative gesture and affect), accepted as universal in the whole spectrum and long-lasting, even in adult's life (Rapin and Dunn, 1997, 2003; Lord & Paul, 2005); ii) impairment regarding semantics i.e the linguistic meaning of utterances and bounds established between words/utterances and what they do/may represent (Rapin & Dunn, 1997, 2003 ; Boucher, 2003).

Overlapping with SLI?

Language was compared in autistic and SLI subjects. In the SLI condition, children fail to develop spoken language, despite adequate hearing, intelligence or physical condition. SLI, that excludes PDD, is defined as a specific and pure impairment (DSM IV), encompassing a broad range of deficits regarding phonology, vocabulary, syntax, semantics and pragmatics. Rapin and Allen (1983) established the following classification: i) mixed receptive/expressive disorders, with comprehension and expression impairment, including verbal-auditory agnosia (word deafness) and phonologic-syntactic syndrome ; ii) expressive disorders, with spared comprehension, including verbal apraxia and phonologic disorder; iii) higher order disorders, with spared articulation and phonology but disordered pragmatics and lexical-syntactic organization. This last category is controversial. Some authors consider it as a part of ASD continuum, while others refuse the systematic equation between pragmatic difficulties and ASD, and identify intermediate characteristics (Bishop & Frazier-Norbury, 2002). Some studies concluded that the *structural disorders* observed in some AD children are similar to those of SLI (especially regarding phonology and syntax), and suggested overlaps between the disorders, hypothesizing a common phenotype (Bartak et al, 1975,1977; Kjelgaard & Tager-Flusberg, 2001 ; Bishop, 2003 ; Tager-Flusberg, 2006).

Conversely, studies focusing on non-word repetition, considered as a psycholinguistic marker of SLI (Bishop et al, 1996; Conti-Ramsden et al, 2001), found in AD and SLI similarities regarding syntactic reception and expression, but differences in speech motor skills, verbal short-term memory, and error types in non-word repetition (Whitehouse & al, 2008). They thus rejected the hypothesis of a SLI subtype in Autism. Some studies considering genetics suggest that the two conditions may be related (Folstein & Mankoski, 2000 ; Vernes et al, 2008), although others discuss an overlapping etiology and “phenomimicry”, suggesting that language impairment in ASD may result from risk factors and differs from that of SLI (Bishop, 2003, 2010). Moreover, although studies regarding ASD and SLI language and communication in first-relatives are consistent with the hypothesis that both disorders are highly inheritable, they do not support the hypothesis of one shared phenotype, but rather suggest that language deficits in ASD and SLI have different origins (Whitehouse et al, 2007; Lindgren et al; 2009). A recent fMRI study found that SLI and Autistic children with Language Impairment (ALI) compared to normal controls showed a smaller anterior vermis, an asymmetry reversal in posterior cerebellum (lobule VIIIA) and Broca's area (Inferior Frontal Gyrus), which suggests abnormalities in the circuits that manage motor control and processing of language, cognition, working memory and attention. They also observed that cerebellar white matter was significantly larger in ALI than in SLI, and regional volume differences between ALI and SLI groups in right hemisphere VIIA Crus I, suggesting similarities but also

important language related developmental differences (Hodge et al, 2010).

Prosody in PDD

Prosody concerns the supra-segmental properties of the speech signal that modulate and enhance its meaning. It supports expressive language at several communication levels, i.e., grammatical, pragmatic and emotional (McCann & Peppé, 2003). Abnormal prosody was identified as a core feature of individuals with autism (Kanner, 1943). The observed prosodic differences include monotonic or machine-like intonation, aberrant stress patterns, deficits in pitch and intensity control and a “concerned” voice quality. These inappropriate patterns related to communication/sociability ratings tend to persist over time even while other language skills improve (Paul et al, 2005a, 2005b). Many studies have tried to define the prosodic features in patients with Autism Spectrum Disorder (ASD) (for a review see McCann & Peppé, 2003). With regards to intonation contours production and intonation contours imitation tasks, results are contradictory. In a reading-aloud task, Fosnot and Jun (1999) found that AD children did not differentiate questions and statements; all utterances sounded like statements. In an imitation condition task, AD children better performed. The authors concluded that AD subjects could produce intonation contours although they do not use them or understand their communicative value. They also observed a correlation between intonation imitation skills and autism severity, which suggests that the ability to reproduce intonation contours could be an index of autism severity. Paul et al. (2005a) found no difference between AD and TD children in the use of intonation to distinguish questions and statements. McCann et al (2007) observed a tendency in AD subjects to utter a sentence sounding like a question when a statement was appropriate. Le Normand et al. (2008) found that children with AD produced more words with flat contours than typically developing children. Paul et al. (2008) documented the abilities to reproduce stress in a nonsense syllable imitation task of an ASD group that included patients with high-functioning autism, Asperger's syndrome and PDDNOS. Perceptual ratings and instrumental measures revealed small but significant differences between ASD and typical speakers.

Prosody in SLI

Intonation was poorly studied in children with SLI (Well & Peppé, 2003). Some researchers hypothesized that intonation provides reliable cues to grammatical structure by referring to the theory of phonological bootstrapping (Morgan & Demuth, 1996), which claims that prosodic processing of spoken language allows children to identify and then acquire grammatical structures as inputs. Consequently, difficulties in the processing of prosodic feature such as intonation and rhythm may generate language difficulties (Weinert, 2000). While some studies concluded that SLI

patients do not have significant intonation deficits and that intonation is independent of both morphosyntactic and segmental phonological impairments (Snow, 1998a, 1998b; Marshall et al, 2009), some others have shown small but significant deficits (Wells & Peppé, 2003 ; Hargrove & Sheran, 1989 ; Samuelsson et al, 2003). With regards to intonation contours production, Wells and Peppé (2003) found that SLI children produced less congruent contours than typically developing children. The authors hypothesized that SLI children understand the pragmatic context but fail to select the corresponding contour. Concerning intonation imitation tasks, the results seem contradictory. Van der Meulen et al. (1997) and Wells and Peppé (2003) found that SLI children were less able to imitate prosodic features. Several interpretations were proposed: (i) the weakness was due to the task itself rather than to a true prosodic impairment (Van der Meulen & al, 1997) ; (ii) a failure in working memory was more involved than prosodic skills (Van der Meulen & al, 1997) ; and iii) deficits in intonation production at the phonetic level were sufficient to explain the failure to imitate prosodic features (Wells & Peppé, 2003). Conversely, Snow (1998b) reported that children with SLI showed a typical use of falling tones and Marshall et al. (2009) did not find any difference in the ability to imitate intonation contours between SLI and typically developing children.

Aims of the study

Most studies aimed to determine whether AD or SLI children's language skills differed or shared an equivalent phenotype, but really few compared AD, PDDNOS and SLI. Mayes & al, (1993) tried to define clinical features that differentiate PDDNOS from both AD and SLI. They found that PDDNOS differed from AD children on items related to the degree of socialization and relatedness. Conversely, they exhibited difficulties with relatedness and needed routines more than SLI. Ramberg et al (1996) compared language and pragmatics functions in AS, High Functioning Autism (HFA), Deficits in Attention, Motor control and Perception (DAMP) and Speech and Language Disordered (SLD) groups. This study clearly discriminated DAMP and SLD from AS and HFA. Last, Allen et al (2001) compared PDDNOS verbal and adaptive skills in PDDNOS, AD and Developmental Language Disordered groups (DLD). Their findings indicated that PDDNOS did not differ from either DLD or AD children in verbal or adaptive skills, obtaining intermediate scores. Nor did they differ from AD regarding maladaptive behaviors, although the two groups had significantly more behaviors of this type than the DLD group. Regarding prosodic skills, studies sought to determine if they AD or SLI differed from those of typically developing children, but rarely focused on differences between diagnostic categories.

The aim of the present study is (i) to document language profiles in AD, PDD-NOS and SLI (phonology, vocabulary, syntax, pragmatics, prosody) and (ii) to determine language differential

markers of pathology. To avoid, previous methodological bias, we first carefully matched our three groups of patients for age, sex and academic skills; and second, we used an automatic intonation recognition algorithm, computed for the current study (Ringeval et al, in press) to assess prosody during a prosodic imitation task.

METHOD

Subjects (Table 1)

Thirty-five monolingual subjects with communicative verbal skills were recruited in two University departments of child and adolescent psychiatry located in Paris, France (Université Pierre et Marie Curie/Pitié-Salpêtrière Hospital and Université René Descartes/Necker Hospital, respectively). They consulted for pervasive developmental disorders (PDD) and specific learning impairments, which were diagnosed as Autistic Disorder (AD), Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS) and Specific Language Impairment (SLI), according to DSM IV criteria (APA, 2002). In the three groups, the age range was 6-18, the sex ratio 70/80% male versus 30/20% female. Socio-demographic and clinical characteristics of the subjects are summarized in table 1.

INSERT TABLE 1 ABOUT HERE

AD and PDD NOS groups were assigned from patients' scores on Autism Diagnostic Interview-Revised (ADI-R, Lord & al, 1994) and Children Autism Rating Scale (CARS, Shopler et al, 1980). The psychiatric assessments and parental interviews were conducted by four child-psychiatrists specialized in autism (JX, DP, DC, LR). To give a developmental view of the older patients with autism and PDD-NOS, the ADI-R diagnosis at age 5 was reported. In 3 subjects, diagnosis had evolved from age 5, from AD to PDD NOS. Of note, all PDD-NOS also fulfilled diagnostic criteria for Multiple Complex Developmental Disorder (Van der Gaag et al., 1995; Buitelaar & Van der Gaag, 1998), a research diagnosis used to limit PDD-NOS heterogeneity and improve its stability overtime (Rondeau et al., in press). As expected, current ADI-R and CARS scores were significantly higher in subjects with AD than with PDD NOS (*Mann-Whitney test*: $U = 80.50$, $p = 0.002$; $U = 112.00$, $p = <0.0001$, respectively). SLI subjects received a formal diagnosis of SLI by Speech Pathologists and Child Psychiatrists specialized in language impairments (DC, LR), according to the DSM IV criteria for expressive language disorder. Additionally, all of them fulfilled criteria for Mixed Phonologic-Syntactic Deficit Disorder, according to Rapin and Allen's classification for Developmental Dysphasia (Rapin & Allen, 1983). This syndrome includes poor articulation skills and ungrammatical utterances at expressive level, and better comprehension skills (in regard to language production), although they were never adequate to age.

Language assessment

Oral language was assessed using 5 tasks of the ELO Battery (*Evaluation du Langage Oral: Assessment of Oral Language*; Khomsi, 2001), i.e., Receptive Vocabulary, Expressive Vocabulary, Word Repetition, Sentence Understanding, Sentence Completion. ELO is dedicated to children 3-11 years old. Although many subjects of our study were older than 11, their oral language difficulties did not allow using other tests because of important floor-effect. Consequently, we adjusted the scoring system and determined severity levels (see below). For most subjects, all tasks were administered in one 30-45 minutes session. For some autistic subjects, 2, 3 or 4 sessions were necessary to complete all tasks.

Receptive Vocabulary

This task containing 20 items requires word comprehension. The examiner gives the patient a picture booklet, and tells him: «*Show me the picture in which there is a ...* ». Subject has to select among 4 pictures the one corresponding to the uttered word. Each correct pointing gives one point and maximal score is 20.

Expressive Vocabulary

This task containing 50 items requires picture naming. The examiner gives the patient a booklet comprising object pictures and asks him «*What is this?*», then «*What is he/she doing ?*» for the 10 last pictures, showing actions. Each correct answer gives one point, maximal mark for objects is 20 for children from 3 to 6, 32 from 6 to 8, and 50 for children above 9.

Word Repetition

This task comprising 2 series of 16 words requires verbal encoding and decoding. The first series contains - disyllabic words, with few consonants groups. The second contains longer words with many consonants groups, which allows observing any phonological disorder. The examiner says «*Now, you are going to repeat exactly what I say. Listen carefully, I won't repeat*». Then patient repeats the 32 words, maximal score is 32.

Sentence Understanding

Oral sentence understanding is assessed with picture designation. There are 32 sentences, 16 simple and 16 more complex requiring accurate grammatical skills. The examiner gives the patient a 34 pages booklet, in which each page contains 4 pictures. There are 2 demonstration items, and instruction is as follows: «*Show me the picture that means*». In case of error, a second choice is offered «*Listen carefully, and show me the picture exactly corresponding to what I say*». Each

correct answer gives one point, and 2 sub-scores may then be calculated: (i) *Immediate understanding*, corresponding to addition of items, simple and complex, succeeded on first trial. Maximal score is 32. (ii) *Global understanding*, corresponding to addition of immediate understanding score and items succeeded on the second trial. Maximal score is 32.

Sentence Completion

In this task, the subject is asked to complete sentences, whose beginning is uttered by the examiner. The patient is given a 28 pages booklet, and each page contains 2 pictures. The examiner comments the first picture, and the patient has to complete the second one that implicates a grammatical variation (plural, negation, passive voice, conjugation...) «*Here, the boy is running, there the boy... (is not running)*». There are 3 demonstration items followed by 25 items, each correct answer gives one point and maximal score is 25. Errors may be categorized as (i) *Linguistic errors*, corresponding to grammatical or syntactic errors, reflecting delayed language skills; and (ii) *Pragmatic errors*, when linguistic or pragmatic context is not taken into account.

Scoring system procedure

We first referred to the means and standard deviations of each task. But due to our population particularities, oral language abilities differed widely from those expected at chronological age. Standard deviations were extremely high and reached floor effect. To adjust the scoring system to all subjects including those older than 11 years (the maximum age limit of the test), we determined for each subject the corresponding age for each score, then we calculated the discrepancy between this «verbal age» and the chronological age. The difference was converted into severity levels using a 5 level Likert-scale, i.e., 0 standing for expected level at chronological age, 1 standing for 1 year deviation from expected level at chronological age, 2 for 2 years deviation, 3 for 3 years deviation, and 4 standing for more than 3 years deviation. As the ELO Battery test is not standardized for subjects older than 11 years, we expected for them the 11-year level. In order to validate the procedure, we calculated Pearson's correlations between raw scores and severity levels. All r^2 coefficients ranged from 0.47 to 1 ($p < 0.0001$), except for word repetition ($r^2 = 0.27$, $P = 0.001$).

Prosodic assessment

Our main goal was to compare children's abilities to reproduce different kind of intonation sentences. In order to facilitate the reproduction and to avoid undue cognitive demand, the sentences were phonetically easy and relatively short. Children were asked to repeat sentences that were randomly presented. We used 4 intonation groups (or prosodic contours) in this study: descending (e.g. "David ate a croissant."); falling (e.g. "Can you go to my office?"); floating (e.g.

“Anna will come with you.”); and rising (e.g. “A croissant?”). Each intonation group includes 7 sentences that ensure different phonetic contexts and thus diversity in prosodic contours (detailed list is given in Ringeval et al, in press). Children were recorded in their usual environment. During recordings, they were asked to repeat exactly the sentences they heard, even if they did not catch one or several words. If the children reproduced the prosodic contours of the sentences in an exaggerated way, or if they showed difficulties, sentences were replayed several times.

As many sources of perturbation appeared during the recordings (e.g. false-starts, repetitions or noise from the environment) we preferred manual based speech segmentation. An automatic detector could hardly handle all perturbations found in the recordings. The sentences reproduced by the children were thus manually segmented and post-processed to keep only those that had complete prosodic contour (i.e. whatever the pronounced words). Nearly 900 sentences were collected from the recordings.

To assess prosodic performance of the children by group, we used an automatic classifier developed to recognize prosodic contour of short sentences taken from the same list. The classifier used both static and dynamic approaches that were fused to provide best recognition scores (see scheme in figure 1 A). Computational characteristics of the classifier are given in detailed in Ringeval et al (in press). Performance of the best fusion configuration on a group of 70 typical developing children matched for age and sex (1 patient for 2 controls) recruited in elementary schools was as follow: descending 0.64, falling 0.55, floating 0.72, rising 0.95. Given that performance of the classifier was below 0.6 for falling sentences, the method was considered not valid for classifying falling sentences in pathological groups.

INSERT FIGURE 1 ABOUT HERE

Statistical analysis

Statistics were done with R statistical software. To compare groups' severity levels in each ELO task, we used one-way analysis of variance followed by *post hoc* analysis. Then, Spearman non-parametric method was used within each group, to test the correlation between raw scores in language tasks, and chronological age. To compare group sentence duration and prosodic performance in the intonation imitation task, we also used a one-way analysis of variance followed by *post hoc* analysis.

RESULTS

Receptive Vocabulary, Expressive Vocabulary and Word repetition

Regarding vocabulary tasks (Table 1), no significant differences were found between groups' mean severity levels: $K(2, 34) = 1.41$, $p = 0.5$ for Receptive task, $K(2,34) = 1.77$, $p = 0.41$ for Expressive task. The 3 groups were similarly delayed (1 to 2 years). Word repetition task showed

impaired phonological encoding skills in the 3 groups, with an average delay of 3 years: $K(2,34) = 0.41, p = 0.81$.

Sentence Understanding

All groups were delayed in immediate understanding (Table 1), and strong significant group differences were observed: $K(2,34) = 14.88, p = 0.0006$. *Post hoc* analyses showed significant differences between AD and SLI groups ($p = 0.001$), and between AD and PDD NOS ($p = 0.005$). However, no differences were found between PDD NOS and SLI groups ($p = 0.08$).

As observed in immediate understanding, all groups were delayed in global understanding and did not benefit from a second trial (Table 1). Again, strong significant differences appeared between groups: $K(2,34) = 16.58, P = 0.0003$. *Post hoc* analysis showed significant differences between AD and SLI groups ($P = 0.0001$), AD and PDD NOS groups ($P = 0.001$), but no differences between SLI and PDD NOS groups ($P = 0.127$).

Sentence Completion

All groups had delayed expressive skills as evidenced by correct sentence scores. Mean severity levels ranged from 1.5 for SLI group, to 2.3 for PDD NOS and 3.2 for AD, with an average delay of 2 years. A significant difference appeared between groups: $K(2,34) = 6.48, P = 0.039$. *Post hoc* analyses showed significant difference between AD and SLI groups, not between AD and PDD NOS groups, nor between PDD NOS and SLI groups. In all groups, the number of linguistic errors was above the expected range according to chronological age, with an average delay of 2 years. No significant differences appeared between groups: $K(2,34) = 1.78, P = 0.41$. In contrast, regarding pragmatic errors, significant differences were found between groups: $K(2,34) = 18.75, P = 0.0001$. SLI subjects obtained scores in the average of children of the same age, whereas AD subjects and PDD NOS committed numerous pragmatic errors. *Post hoc* analyses showed significant differences between AD and SLI groups ($p < 0.0001$), PDD NOS and SLI groups ($p = 0.001$), and also between PDD NOS and Autistic groups ($p = 0.044$).

Correlations study

Correlations between raw scores in language tasks and chronological age, within each group

In the AD group, no correlations were found between raw scores in language tasks and chronological age, neither in the PDD NOS group, except for pragmatic errors: $r_s(10) = 0.72, p = 0.02$. Unlike the two previous groups, all language tasks' raw scores were strongly and positively correlated to chronological age in the SLI group (Table 2), except for Linguistic Errors: $r_s(12) = -0.90, p < 0.0001$ and Pragmatic errors: $r_s(12) = -0.61, p = 0.04$, where correlations were negative.

INSERT TABLE 2 ABOUT HERE

Correlations within raw scores in language tasks, within each group

In the AD group, few correlations appeared within tasks (Table 3, top). Receptive Vocabulary raw scores were correlated to Expressive Vocabulary: $r_s(12) = 0.74, p = 0.03$, Word Repetition: $r_s(12) = 0.76, p = 0.02$ and Correct Sentences: $r_s(12) = 0.85, p = 0.006$. Expressive Vocabulary was also correlated to Word Repetition: $r_s(12) = 0.74, p = 0.03$. More correlations were observed in the PDD NOS group, as most of the tasks' scores were correlated within them, p values ranging from 0.04 to <0.0001 . However, Receptive Vocabulary, Linguistic Errors and Pragmatic Errors were not correlated to any other component (Table 3, middle). In the SLI group, all raw scores were strongly and positively correlated within them, p values ranging from <0.0001 to 0.013. Unlike previous groups, even Linguistic Errors were correlated, though negatively, to all the other language components (Table 3, bottom). Pragmatic Errors were here negatively correlated to Immediate Understanding: $r_s(12) = -0.61, p = 0.04$ and to Global Understanding $r_s(12) = -0.60, p = 0.045$.

INSERT TABLE 3 ABOUT HERE

Performances in the intonation imitation task

Compared to the reference sentences, sentence duration was conserved for all intonation groups when reproduced by TD children. In contrast, durations of pathological group intonation sentences were strongly different from those of TD children (Table 4, top). Values were lengthened by 30 % for the three first groups of intonation sentences and by more than 60 % for the "Rising" contour. Moreover, the duration of SLI children's sentences intonation was significantly longer than those of AD and PDD-NOS groups, except for "Rising" intonation.

Characteristics of the automatized classifier are shown in figure 1. For TD children, best recognition scores were obtained with a major contribution of the static classifier ($\alpha=0.7$) that is based on global statistics of features of prosody (Figure 1B). In contrast, in children with language impairments (LIC), best recognition scores were obtained with a major contribution of the dynamic classifier ($1-\alpha=0.9$) allowing modelling the temporal structure of the features (Figure 1C). Abilities in reproducing prosodic contours as assessed by the automatized classifiers with the best fusion for each group are presented in table 4 (bottom). All pathological groups recognition scores were close to those of TD children and similar between LIC's groups for "Descending" intonation. In contrast, all other intonations were significantly different between TD and all pathological groups. However, SLI and TD children had very high recognition rate for the "Rising" intonation while both AD and PDD-NOS performed significantly much lower.

INSERT TABLE 4 ABOUT HERE

DISCUSSION

Summary of the current results

The aim of the study was to document language profiles in French-speaking SLI, PDDNOS, AD and to determine differential markers of pathology. At the lexical and phonological levels we observed a similarity between the 3 groups in vocabulary and word repetition tasks, with a respective average delay of 1-2 years and 3 years. *Vocabulary and phonology thus do not appear as differential but rather general markers of developmental pathology. Lexical and phonological skills are vulnerable, whatever the children's impairment.* Regarding syntactic skills, at the receptive level, the groups were delayed in sentence understanding; significant differences were observed between AD and SLI groups, AD and PDD NOS, but not between PDD NOS and SLI groups. At the expressive level, the 3 groups were similarly delayed, producing few correct sentences. Significant differences were observed between AD and SLI groups, but not between AD and PDD NOS, nor between PDD NOS and SLI groups. Regarding expressive errors patterns, the three groups produced similar linguistic errors above the expected range. Our findings confirm other studies that observed poor comprehension (Boucher, 2003; Rapin and Allen, 1997; Bartak et al, 1977; Tager-Flusberg, 1981), immature syntax and prevalence of syntactic errors in PDD (Kjelgaard & Tager-Flusberg, 2001). In our study, AD performed lower than PDD-NOS at the receptive level. *Expressive syntax thus appears as a differential marker between AD and the two other groups.* Regarding pragmatic errors, significant differences were found between the groups. SLI subjects obtained expected scores, AD subjects and PDD NOS were deficient, and PDD NOS performed significantly better than AD. This finding is of interest as PDD-NOS is often included in PDD, and not considered as a category. Inasmuch as AD appeared pragmatically more deficient than PDD-NOS. *Pragmatic skills could be considered as a differential marker between AD and the other groups, notably PDD-NOS.*

At the prosodic level, fusion parameters of the classifier for best recognition scores were very different between TD children and LIC, meaning that the way to achieve a certain quality of prosody was not similar in TD and LIC, whatever the pathological subgroup. In terms of intonation imitation, the pathological groups and the TD children performed similarly for the "Descending" intonation pattern. In contrast, for the "Rising" intonation pattern, SLI and TD children had very high recognition rates, whereas both AD and PDD-NOS performed significantly much lower. *This suggests that one feature of prosody, the "rising intonation", is a differential marker of psychopathology, allowing separating SLI and PDD groups.* Such finding is in accordance with studies showing that SLI patients do not have significant intonation deficits and that intonation is independent of both morphosyntactic and segmental phonological impairments (Snow, 1998a, 1998b; Marshall et al, 2009).

PDD-NOS: Developmental delay or pervasive development?

In children matched for age, sex and academic level, language skills appear as differently spared and affected in SLI, PDD-NOS and AD groups. The AD group is the most deficient, presenting difficulties at the lexical, syntactic, pragmatic and prosodic levels. The SLI group is comparable to normal children at the pragmatic level and some aspects of the prosodic level. PDD-NOS and SLI groups perform similarly at the lexical and syntactic levels, but not at the pragmatic and prosodic ones. The PDD-NOS group appears as intermediate, close to SLI in lexical and syntactic skills, poorer than SLI as but better than AD in pragmatic and prosodic skills. The correlation study also supported an intermediate position for PDD-NOS. Indeed, in SLI, we found high correlation rates between language scores and chronological age, as well as between scores in the different language tasks. This profile confirms the view that SLI is a developmental language disorder and supports a delayed profile as intellectual disability for general cognitive skills. In contrast, we found no correlations in AD, neither between language scores and chronological age, nor between language scores in the different language task, suggesting a deviant or pervasive development of language skills in autism. The correlation between language skills with age for SLI and the lack of correlation with age for AD was also found in an automated vocal analysis of naturalistic recordings from children with autism, language delay, and typical development (Oller et al., 2010). In our study, PDD-NOS, shared with SLI some correlations between the scores in the different language tasks, showing that development of language followed some kind of hierarchy. However, like AD, PDD-NOS showed no correlation between language scores and chronological age. Therefore, as regard the language dimension, PDD-NOS showed some form of developmental delay, but also some form of pervasive development. The language deficit of the AD group appears as more global than those of the two other groups, which suggests large cognitive abnormalities involving language disturbances, as shown by a recent fMRI study (Hodge et al, 2010).

Implication for DSM-5

Theoretically, our study does not support the hypothesis of an overlapping and common phenotype between AD and SLI (Bartak et al, 1975, 1977; Kjelgaard & Tager-Flusberg, 2001; Bishop, 2003; Tager-Flusberg, 2006) and tends to reject, as did other authors (Bishop et al, 1996; Conti-Ramsden et al, 2001), the hypothesis that SLI could be a subtype of PDD. Taken together, our findings evidence the clinical necessity to assess every language domain in all pathological groups, in order to refine diagnosis criteria and specify language remediation axis. Focusing on syntactic, pragmatic and prosodic markers could be of strong differential interest. The possibility to use engineering approach to automatically assess prosodic skills is of high interest as it has been shown

to be able to distinguish pathological subjects with language impairment in naturalistic setting (Warren et al., 2010; Oller et al., 2010), but also in an experimental imitation task (Ringeval et al., in press).

With respect to the current proposed revision of DSMIV-R criteria for ASD including merging the three subtypes, AD, AS and PDD-NOS, into one category, namely Autism Spectrum Disorder (ASD), Rondeau et al (in press) studied stability over time, and showed the lack of support for reliably distinguishing PDD-NOS as a diagnostic entity. They clearly demonstrated the heterogeneity of the PDD-NOS group which would impact the predictive validity of the proposed DSM-V entity. PDD-NOS would be conceived as corresponding to a group of heterogeneous pathological conditions including prodromic forms of later AD, remitted or less severe forms of AD, and developmental delays in interaction and communication. The international clinical and research consensus on the robustness of AD as defined by currently more stringent DSM criteria, would be lost. International communication and comparison between interventions will be jeopardized.

The DSM5 expert Committee does not consider language as a differential marker (<http://www.dsm5.org/ProposedRevisions/Pages/proposedrevision.aspx?rid=94#>). Our study strongly suggests the pertinence of distinguishing SLI, AD and PDD-NOS inasmuch as the language developmental pattern, when finely assessed, appears as specifically altered and spared in the three groups. To limit PDD-NOS heterogeneity (Towbin, 2005) and stability (Rondeau et al., in press), another view, would be to develop appropriate clinical algorithm to individuate PDD-NOS subtypes as proposed earlier with the concept of Multiplex Developmental Disorder (Cohen et al, 1997; Van der Gaag, 1995). However, if the enlarged “ASD” diagnosis is adopted in DSM-5, it could be important to recommend a dimensional clinical approach of the patients including language as it may have major implication regarding treatment and remediation.

Conclusion

Our findings support the hypothesis that language skills and development in AD and SLI rely on different mechanisms, while PDDNOS show an intermediate profile, sharing some characteristics of both AD and SLI. They also suggest that expressive syntax, pragmatic skills and some intonation features could be considered as language differential markers of pathology.

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Table 1. Socio-demographic, clinical and language characteristics of the subjects			
	AD (N=12)	PDD-NOS (N=10)	SLI (N=12)
Age, mean (\pm SD), year	9.75 (\pm 3.5)	9.83 (\pm 2.17)	9.17 (\pm 3.9)
Male – Female	10 – 2	9 – 1	9 – 3
ADI-R, current, mean (\pm SD)			
Social impairment score	21.1 (\pm 5.8)	12.7 (\pm 7.8)	Not relevant
Communication score	19.3 (\pm 5.2)	8.5 (\pm 6.4)	Not relevant
Repetitive interest score	6.4 (\pm 2.4)	2.0 (\pm 1.6)	Not relevant
Total score	50.7 (\pm 12.8)	25.7 (\pm 15.4)	Not relevant
CARS score	33.2 (\pm 15.4)	22.3 (\pm 5.4)	Not relevant
ADI-R, 5 year-old			
Autism	N=12	N=3	Not relevant
PDD-NOS	N=0	N=7	Not relevant
Cognitive Level			
WISC3/WPPSI			
VIQ	50 (\pm 8.3)	85 (\pm 14.4)	71.3 (\pm 11.7)
PIQ	77 (\pm 16.3)	76.8 (\pm 10.5)	95.4 (\pm 14.5)
WISC 4			
ICV		79.8 (\pm 26.2)	96 (\pm 19.8)
IRP		83 (\pm 25.1)	95 (\pm 22.6)
IMT		75 (\pm 20.1)	77.5 (\pm 6.4)
IVT		83.5 (\pm 22.6)	99 (\pm 18.4)
ELO tasks: mean (\pm SD)			
Receptive Vocabulary	2.4 (\pm 1.6)	1.9 (\pm 1.5)	1.9 (\pm 1.0)
Expressive Vocabulary	2.0 (\pm 1.8)	1.2 (\pm 1.8)	1.4 (\pm 1.1)
Word Repetition	2.9 (\pm 1.5)	2.7 (\pm 1.4)	3.5 (\pm 0.7)
Immediate Understanding	3.4 (\pm 1.2)	1.8 (\pm 1.5)	0.8 (\pm 0.9)
Global Understanding	3.3 (\pm 1.1)	1.4 (\pm 1.6)	0.6 (\pm 0.9)
Correct Sentences	3.2 (\pm 1.3)	2.3 (\pm 1.3)	1.7 (\pm 1.5)
Linguistic Errors	2.7 (\pm 1.4)	2.1 (\pm 1.5)	1.8 (\pm 1.8)
Pragmatic Errors	3.5 (\pm 1.0)	2.3 (\pm 2.0)	0.3 (\pm 0.9)

AD= Autistic Disorder; PDD-NOS= Pervasive Developmental Disorder-Not Otherwise Specified; SLI= Specific Language Impairment; SD= Standard Deviation; ADI-R=Autism Diagnostic Interview-Revised; CARS= Children Autism Rating Scale.

Table 2. Spearman rank correlations table, between raw scores in language tasks and chronological age within each group (AD, PDD-NOS and SLI patients)

<i>ELO tasks</i>	AD age Spearman's ρ	PDD NOS age Spearman's ρ	SLI age Spearman's ρ (p)
<i>Receptive Vocabulary</i>	-0.05	-0.21	0.84***
<i>Expressive Vocabulary</i>	-0.03	-0.08	0.90***
<i>Word Repetition</i>	0.25	-0.07	0.88***
<i>Immediate Understanding</i>	-0.17	0.24	0.95***
<i>Global Understanding</i>	0.38	0.25	0.87***
<i>Correct Sentences</i>	-0.35	0.08	0.94***
<i>Linguistic Errors</i>	0.22	-0.11	-0.90***
<i>Pragmatic Errors</i>	0.14	0.72*	-0.61*

* $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

AD= Autistic Disorder; PDD NOS= Pervasive Developmental Disorder Not Otherwise Specified;
SLI= Specific Language Impairment

Table 3. Spearman rank correlations table within raw scores in language tasks for AD, PDD-NOS and SLI patients

<i>Autism Disorder: r_s</i>								
	<i>Rec.Voc</i>	<i>Exp.Voc</i>	<i>W.Rep</i>	<i>Imm.Und</i>	<i>Glo.Und</i>	<i>Corr.Sen</i>	<i>Ling.Err</i>	<i>Prag.Err</i>
<i>Rec.Voc</i>	1	0.74*	0.76*	0.24	0.48	0.85**	-0.04	-0.51
<i>Exp.Voc</i>	0.74*	1	0.74*	0.50	0.36	0.62	0.01	0.0
<i>W.Rep</i>	0.76*	0.74*	1	-0.15	0.13	0.50	-0.09	-0.10
<i>Imm.Und</i>	0.24	0.50	-0.15	1	0.59	0.39	0.03	0.01
<i>Glo.Und</i>	0.48	0.36	0.13	0.59	1	0.28	0.15	-0.37
<i>Corr.Sen</i>	0.85**	0.62	0.50	0.39	0.28	1	-0.13	-0.43
<i>Ling.Err</i>	-0.04	0.01	-0.09	0.03	0.15	-0.13	1	-0.34
<i>Prag.Err</i>	-0.51	0.004	-0.10	0.01	-0.37	-0.43	-0.34	1
<i>Pervasive Developmental Disorder-NOS: $r_s(p)$</i>								
	<i>Rec.Voc</i>	<i>Exp.Voc</i>	<i>W.Rep</i>	<i>Imm.Und</i>	<i>Glo.Und</i>	<i>Corr.Sen</i>	<i>Ling.Err</i>	<i>Prag.Err</i>
<i>Rec.Voc</i>	1	0.26	-0.11	0.20	0.20	0.25	-0.33	-0.31
<i>Exp.Voc</i>	0.26	1	0.71*	0.82**	0.81**	0.67*	-0.36	-0.29
<i>W.Rep</i>	-0.11	0.71*	1	0.83**	0.78**	0.86**	-0.33	-0.30
<i>Imm.Und</i>	0.20	0.82**	0.83**	1	0.96***	0.90***	-0.57	-0.20
<i>Glo.Und</i>	0.20	0.81**	0.78**	0.96***	1	0.90***	-0.53	-0.24
<i>Corr.Sen</i>	0.25	0.67*	0.86**	0.90***	0.90***	1	-0.61	-0.34
<i>Ling.Err</i>	-0.33	-0.36	-0.33	-0.57	-0.53	-0.61	1	0.15
<i>Prag.Err</i>	-0.31	-0.29	-0.30	-0.20	-0.24	-0.34	0.15	1
<i>Specific Language Impairment: $r_s(p)$</i>								
	<i>Rec.Voc</i>	<i>Exp.Voc</i>	<i>W.Rep</i>	<i>Imm.Und</i>	<i>Glo.Und</i>	<i>Corr.Sen</i>	<i>Ling.Err</i>	<i>Prag.Err</i>
<i>Rec.Voc</i>	1	0.85***	0.71*	0.78**	0.73**	0.78**	-0.78**	-0.54
<i>Exp.Voc</i>	0.85***	1	0.88***	0.86***	0.91***	0.93***	-0.89***	-0.39
<i>W.Rep</i>	0.71*	0.88***	1	0.81**	0.78**	0.82**	-0.79**	-0.26
<i>Imm.Und</i>	0.78**	0.86***	0.81**	1	0.90***	0.93***	-0.93***	-0.61*
<i>Glo.Und</i>	0.73*	0.91***	0.78**	0.90***	1	0.94***	-0.90***	-0.60*
<i>Corr.Sen</i>	0.78**	0.93***	0.82**	0.93***	0.94***	1	-0.95***	-0.57
<i>Ling.Err</i>	-0.78**	-0.89***	-0.79**	-0.93***	-0.90***	-0.95***	1	0.47
<i>Prag.Err</i>	-0.54	-0.39	-0.26	-0.61*	-0.60*	-0.57	0.47	1

* $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

AD= Autistic Disorder; PDD NOS= Pervasive Developmental Disorder Not Otherwise Specified; SLI= Specific Language Impairment; r_s = Spearman's rho ; P = P value ; Rec.Voc=Receptive Vocabulary ; Exp.Voc= Expressive Vocabulary ; W.Rep= Word Repetition ; Imm.Und= Immediate Understanding ; Glo. Und= Global Understanding ; Corr.Sen= Correct Sentences ; Ling.Err= Linguistic Errors ; Prag.Err= Pragmatic Errors

Table 4. Sentence duration and prosodic performances of typical developing children and children with AD, PDD-NOS and SLI during the intonation task

Intonation	TD	AD	PDD-NOS	SLI
<i>SENTENCES DURATION (in seconds)</i>				
Descending	1.7 (0.6)	2.2 ^{*T,S} (0.9)	2.2 ^{*T,S} (0.8)	2.4 ^{*T,A,P} (0.9)
Falling	1.3 (1.4)	1.6 ^{*T} (0.6)	1.7 ^{*T} (0.8)	1.8 ^{*T,A,P} (0.8)
Floating	1.6 (0.4)	2.1 ^{*T,S} (0.7)	2.1 ^{*T} (0.5)	2.4 ^{*T,A,P} (1.0)
Rising	0.5 (0.2)	0.9 ^{*T} (0.3)	0.9 ^{*T} (0.3)	0.8 ^{*T} (0.2)
<i>INTONATION RECOGNITION PERFORMANCES</i>				
Descending	64	64	70	63
Floating	72	48 ^{*T}	40 ^{*T}	31 ^{*T}
Rising	95	57 ^{*T,S}	48 ^{*T,S}	81 ^{*T,A,P}
All including falling	77	56 ^{*T}	53 ^{*T}	58 ^{*T}

Statistics of sentences duration are given for both mean (normal style) and standard-deviation (italic style and between parenthesis) values; Performances are given in percentage of recognition performed by the automatic system intonation recognition where a stratified 10-cross-fold validation based approach crossed with the one employed for TD children was used. Intonation's performances do not include those of "Falling" intonation as recognition score in TD was below 0.6 (see method).

*Kruskal-Wallis' significativity level below 5% compared to children's groups, i.e., T, A, P and S; TD (T): typically developing; AD (A): autism disorder; PDD (P): pervasive developmental disorders not-otherwise specified; SLI (S): specific language impairment.

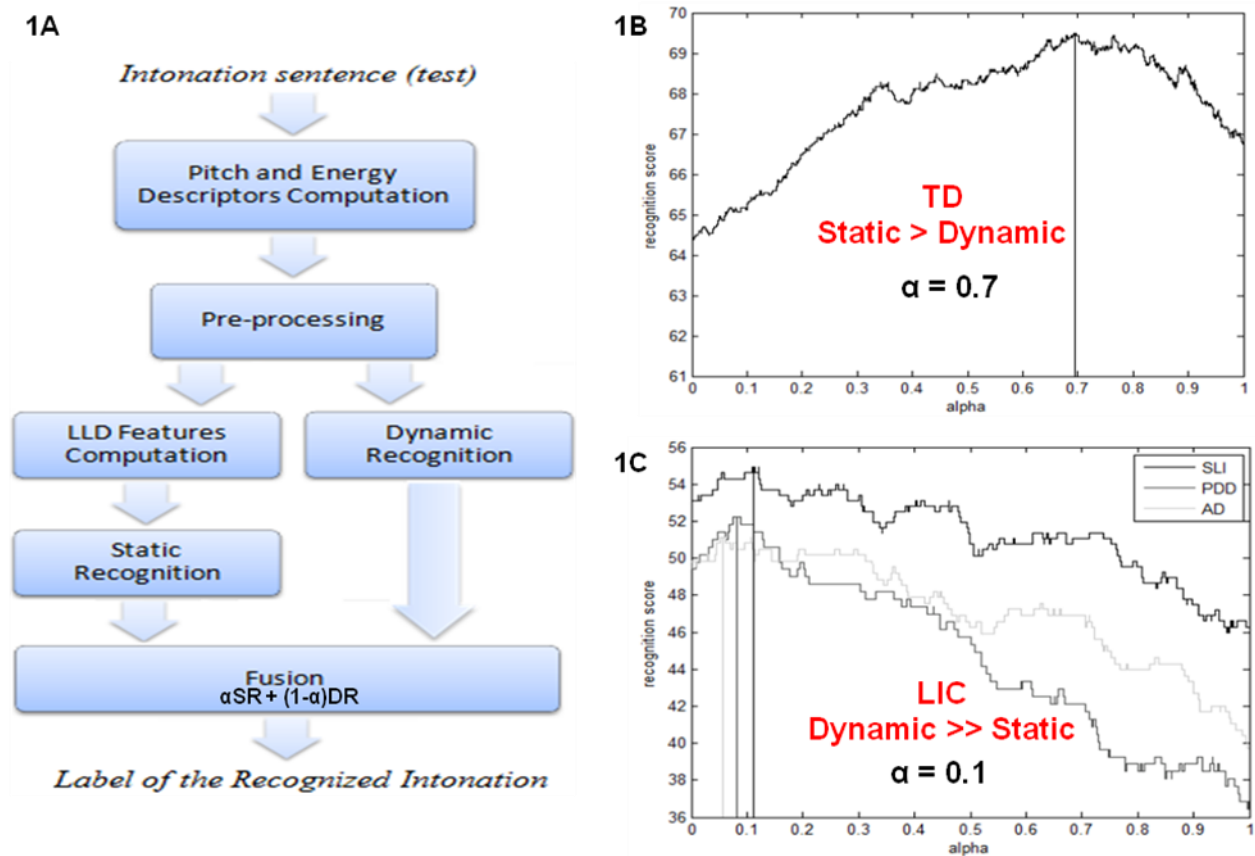


Figure 1. Relative contribution of static classifier and dynamic classifier of prosody for best recognition of intonation sentence according to typical development or language impairment

1A. Scheme of the automatized intonation recognition system. **1B.** Fusion recognition scores as function of weight alpha attributed to both static and dynamic classifiers in typical developing children (TD). **1C.** Fusion recognition scores as function of weight alpha attributed to both static and dynamic classifiers in language impairment children (LIC).

SLI= Specific Language Impairment; PDD=Pervasive Developmental Disorder-NOS; AD=Autism Disorder; LLD=Low-Level Descriptors; SR=Static Recognition; DR= Dynamic Recognition.

(Adapted from Ringeval et al. *IEEE TASP*, in press)