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Neuromotor Development and Language Processing in Developmental Dyspraxia: A Follow-Up Case Study*

Marie-Thérèse Le Normand[1], Laurence Vaivre-Douret[2], Christiane Payan[3], and Henri Cohen[4]
[1]Groupe Hospitalier Pitié-Salpêtrière, [2]INSERM, U483, [3]Hôpital St Vincent de Paul, Paris, France,
[4]Université du Québec, Montreal, Canada

ABSTRACT

A longitudinal study of a child (MV) with developmental verbal dyspraxia was conducted to determine to what extent language development and motor performance in this clinical diagnosis followed a similar course of maturation. Patient MV was observed for two years from the age of 5 years and 6 months. Initially, this young patient exhibited unintelligible and atypical speech production (multiword utterances without consonants), delay in balance and coordination, and impairments in rhythmic tasks; but she was otherwise developing normally with no intellectual impairment or behavioral disorder. MRI scans showed moderately enlarged ventricles, a thin, incompletely myelinated corpus callosum and intact basal ganglia. Two years later, MV's performance was nearly normal only in comprehension aspects of language. In contrast, production aspects of language and speech and neuromotor development showed very little improvement after two years. These observations first suggest that development of receptive and expressive domains within language may be asynchronous, and that the progression of motor control of language appears to follow a parallel course to neuromotor development.

Developmental dyspraxia (DD) is not a singular syndrome. Sensory integration theory, for example, makes a distinction between execution and planning problems in DD (Ayres, 1972a, 1972b). Cermak (1985) also noted that therapists in clinical practice distinguished between children who showed motor planning deficits and those with deficits in the coordination or in the execution of motor tasks. The former appeared to have a general problem in organizing and planning their approach to tasks (i.e., primary planning apraxia), whereas the latter appeared to know how to plan their approach to a particular task but experienced difficulty in executing the task (i.e., executive apraxia).

The literature on neurodevelopment has tended to apply the term *dyspraxia* when referring to disorders in children who demonstrate motor problems that are not due to basic motor impairment, such as infantile hemiplegia or to ''hard'' neurological signs. Dyspraxia is often used interchangeably or synonymously with terms such as clumsiness, sensorimotor dysfunction, or developmental coordination disorder. As a result, different types of developmental motor disorders have not been well defined or consistently described (Cermak, 1985; Cermak, Costers & Drake, 1980; Denckla & Roeltgen, 1992; Henderson, 1987). In some cases, motor deficits are just one component of a general picture of delayed or retarded development. In others, mo-

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Address correspondence to: Dr. Marie-Thérèse Le Normand, Hôpital de la Salpêtrière, Bât. Nlle Pharmacie, 47 Bld de l'Hôpital, 75651 Paris Cedex 13, France. E-mail: lenorman@infobiogen.fr
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tor deficits are not accompanied by a delay in intellectual development but by various academic problems (Henderson, 1987). Finally, some children may evidence only developmental motor deficits (Gubbay, 1979).

In addition to motor deficits, children with DD have also been found to demonstrate language and speech production problems. Few studies, however, have investigated the specific language deficits of children who exhibit both language and other types of motor deficits (Crary, 1984; Crary & Anderson, 1990; Crary, Landness & Towne, 1984; Dodd, Leahy & Hambly, 1989; Le Normand, 1993; Le Normand & Chevrie-Muller, 1991; Le Normand, Konopczynski & Vinter, 1993; Lyytinen & Ahonen, 1988; Schwartz & Regan, 1996).

The influence of biomechanical and motor control constraints on emerging language has been examined by MacNeilage, Hutchison & Lasater (1981) and Kent (1981, 1984, 1992), who pointed out that acquisition of phonology interacts with the acquisition of motor control for speech and that the development of the child's motor facility also depends in part on the need to utter contrasting sequences of speech. Speech production has been described as having two relatively independent stages: a premotor stage during which selection and sequencing of speech segments occur, and a motoric stage that involves the control of movement for production of the segments. MacNeilage et al. (1981) investigated the extent to which this two-stage view could be extended to very early speech development. They reviewed evidence that a child may proceed gradually from a holistic stage of word production to a stage in which there is a separate premotor representation of individual sounds. MacNeilage et al. (1981) suggested early consonantal production preferences as the result only of the motor constraints of the production system and not as a representation in a premotor stage.

The literature on first language, both normal and impaired, has frequently alluded to different levels of processing as an explanation for the processes or ''realization rules'' which characterize the language output of chidren. For example, constraints on articulation, phonological representations (Ingram, 1974, 1976, 1986) and more specifically output lexical representations have been suggested (Menn , 1983) as possible domains of impairment or difficulty. Questions about the processing deficits underlying speech disorder have sometimes been raised (Camarata et Gandour, 1984; Crystal, 1980; Edwards, 1983; Grunwell, 1981; Ingram, 1976; Leonard & Brown, 1984); and preliminary models of the development of speech processing have also been proposed, along with evidence that bears on which and how levels of processing are affected in the disorder. Hewlett's model (1985, 1988) proposes that there is a ''motor programmer'' which receives a perceptual representation from the input lexicon and devises a plan that is then relayed to a motor processing component.

The relationship between language and motor development has usually been discussed primarily as a co-occurring rather than as an interactive maturation. Molfese and Betz (1987) reviewed the literature describing three separate views of language and motor development. One view suggests separate maturation and cerebral lateralization for language and motor processes. Another view considers language and motor development as processes that share overlapping hemisphere sites, allowing for some degree of shared hemisphere lateralization. Molfese and Betz (1987) favor a third view, according to which language and motor development evolve synchronously due to constraints in central nervous system maturation. Language and motor development are ongoing processes and the co-occurrence of these two general classes of behavior is best explained as a set of parallel developments in a developing organism.

Most of the evidence reviewed by Molfese and Betz (1987) compared language production with the development of fine and gross motor skills in the limbs. Much data has been offered describing similarities in speech and limb systems with respect to timing patterns, responses to applied perturbations, and basic kinematic characteristics (Ostry & Cooke, 1987). Although these studies illustrate similarities in the control of movement in the two systems, they do not address whether they influence each other. Examination of how motor control develops in

general, and specifically how the development of speech motor control interacts with the development of language is an important issue for clinical research.

From this perspective, the present study aims to explore the motor deficits underlying different patterns of language output processing in order to provide some empirical evidence concerning the relationship between language and motor development. This follow-up case study is about a child diagnosed with DD. This child showed neither cognitive, hearing nor oromotor deficits but did display a severe impairment of speech production co-occurring with a mild delay in motor tasks. She produced many vowels and almost no consonants when first seen at the age of 5 years and 6 months. This case thus offers a very interesting opportunity to help us better understand the relationship between speech and nonspeech motor ability.

CASE PRESENTATION

The subject in this study, MV, was the youngest of three siblings and was referred at age 5 years 6 months to the Saint Vincent de Paul Hospital for a neurodevelopmental assessment (language and motor evaluation). The history reported by her mother indicated that MV was born without any complications. The mother reported identifying ''speech'' problems quite early on in the development of this child. She also reported that MV was left-handed: this was confirmed by direct observation. MV started walking at 18 months, and had normal hearing as confirmed by an audiological assessment. Moreover, there were no anatomical abnormalities of the oral cavity and simple voluntary movement of the lips, tongue, palate and velar motricity were normally produced, both on command and spontaneously. The mother also added that sentence-like intonation patterns began at around 18 months of age. The first emergent meaningful speech also followed an unusual course of development. The only reduplicated CV syllables found were when the child produced the first three intelligible words (*papa*/daddy, *maman*/mummy, and *bébé*/baby) at about 36 months of age. Since then, her speech has been characterized by very poor intelligibility — essentially due to the absence of consonants in her speech. Nonetheless, her parents learned to recognize her ''sentences'' due to correct syllabic and intonation patterns in relation to context. Although communicative intentions were generally well established, MV was still unable to produce verbal forms in consistent syllabic shapes.

Evaluation at Age 5 Years and 6 Months

Neuromotor assessment

Magnetic resonance imagery revealed moderately enlarged ventricles and a thin incompletely myelinated corpus callosum. The basal ganglia appeared intact. Figure 1 shows the MRI scans. The neurological examination (Amiel-Tison, Njiokiktjien, Vaivre-Douret et al., 1996) focused on cranial neurological signs, passive tone of upper limbs, lower limbs and axis, primitive reflexes and ankle stretch responses. In addition, possible neuroanatomical correlates of brain damage were investigated via magnetic resonance imagery (MRI) of the brain.

At 5 years and 6 months of age, MV showed an imbalance of axial tone with hyperextension; and excessive extension of the lower limbs was limited. Rotulian reflexes were quick. MV also showed a bilateral phasic triceps stretch reflex response to the ankle dorsiflexion maneuver that was more obvious on the right foot than on the left foot. A Babinski reflex was also noticed on the right foot.

Evaluation of neurological tonic laterality revealed that MV behaved as a righthanded hypertonic child with synkinesis of imitation for both the upper and lower limbs. MV appeared as a right-handed child in spontaneous gestures, as in the crossing of arms and ranked at the 50th percentile on Bergès arms/hands tasks and complex gestures (Bergès & Lézine, 1972). She walked with a sustentacular polygonal pattern and had a left foot varus. Walking on a line was more difficult than walking spontaneously. She also jumped like a 2-year-old child (Douret, 1993) and was unable to do bimanual symmetric and alternate pronation-supination movements

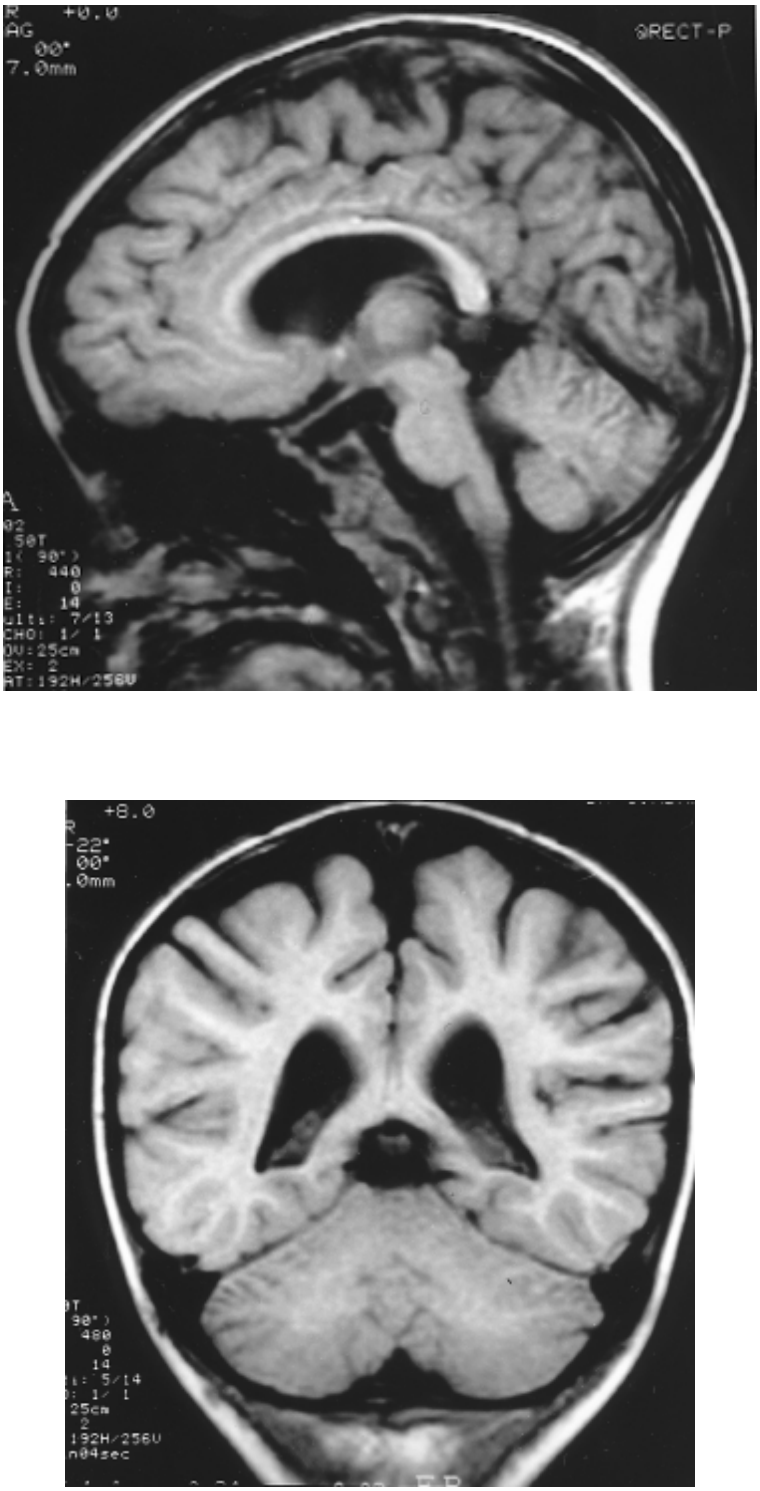

Fig. 1. MRI examinations of case MV at 5 years and 4 months of age. a) Sagittal view showing thin corpus callosum. b) Transverse view showing moderately enlarged ventricular system.

(Ramaeckers & Njiokiktjen, 1991). Overall, dynamic balance was very impaired.

Finally, MV showed hypertonia in her writing posture and an awkward grasp of the pencil. She was able to reproduce a circle and a square but not a triangle. Her scores on the Developmental Test of Visual Motor Integration (Beery, 1982) corresponded to those of children aged 4 years and 4 months.

Language assessment

Cognitive assessment included the Leiter International Performance Scale (LIP; Leiter, 1948) as well as language tasks. MV's I.Q. was 105 and she scored within the normal range for her chronological age on the two auditory memory subtests of the French language examination (Chevrie-Muller, Simon & Decante, 1981). In the rhythmic tapping subtask, however, her performance was 2 *SD* above the mean for her chronological age.

MV was given a battery of six language tests, three of linguistic comprehension and three of linguistic production. Comprehension tasks included a *pointing* task (subject was asked to choose which of a set of images or objects had been named by the experimenter), *understanding of narrative* (subject was required to answer to what, where, who, how, why-type questions in the context of a story, ''The fall in the mud''), and *understanding of prepositions* (representation of space, quantity and partition). The linguistic production tasks included *repetition* (repeating 33 simple words spoken by the experimenter with concurrent visual presentation of the referents), *lexical diversity* (number of different words uttered in a 20 minutes controlled play session) and *grammatical production* (as measured by MLU). Normative data on all these tasks are found in Chevrie-Muller et al. (1981). The measures of lexical diversity and grammatical production were derived from 20 minutes of spontaneous language production, recorded during a standardized play session with Fisher-Price toy figures and objects, replicas of household items and characters (Le Normand, 1986). MV was tested singly in interaction with an adult; and verbal interactions were transcribed directly from the videotape to a Macintosh computer by two speech and language therapists. Transcription was done following Rondal, Bachelet and Pérée (1985) coding; and analysis was done following the Child Language Data Exchange System (CHILDES; MacWhinney & Snow, 1991) convention for automated analysis of language transcripts. CHILDES includes precise guidelines for orthographic transcription and coding (Codes for Human Analysis of Transcripts; CHAT) and programs for analysis (CLAN). In this procedure, the sample is segmented into utterances based on terminal intonation contours and pauses. Inter-rater agreement of transcribed material was above 90%. MV's production of consonants included the labials /p, b, f, v, m/ and the linguals /t, d, k, g, s/š, z/ž, l, r, n/.

Observations showed that MV obtained average scores in all three tasks of comprehension, suggesting that her performance was within the normal range for her chronological age. In contrast, measures of lexical diversity and grammatical production showed that MV scored lower than the normal range of children aged 2 years and 9 months. MV's performance in the simple repetition task revealed poor control of the vocal tract, groping for articulatory postures and inconsistent articulatory output. Her speech errors included phonetic distortions, syllable reduction for multisyllabic words and difficulties with articulatory place change. The percentage of words with correct articulation was 9% (3/33). The words, however, were produced with appropriate intonational contour, suggesting that MV was able to adequately mark syllable phrases (217 Hz < Mean F_0 > 265 Hz ; Min/Max F_0 = 177/333 Hz).

Evaluation at Age 7 Years and 6 Months

Neuromotor assessment

There was no noticeable change after two years.

Language assessment

Performance in comprehension was normal relative to the average performance of a control population with the same chronological age. Production scores, however, revealed that MV's

lexical diversity is still significantly below that of normal 3-year-old French-speaking children (see Le Normand, 1997, for a description of the linguistic norms). MV's grammatical production has somewhat improved over the space of two years but still remains within the range of 3-year-old children. Simple repetition has also improved, and the percentage of words with correct articulations has now reached 30%. The words were also produced with appropriate intonation (231 Hz < Mean F_0 < 295 Hz; Min/Max F_0 = 122/386 Hz). Figures 2 and 3 and Table 1 show the evolution of MV's linguistic performance.

DISCUSSION

The main observation in this follow-up study of a child with developmental dyspraxia is that evolution of specific competencies in language may follow a different course in individuals with

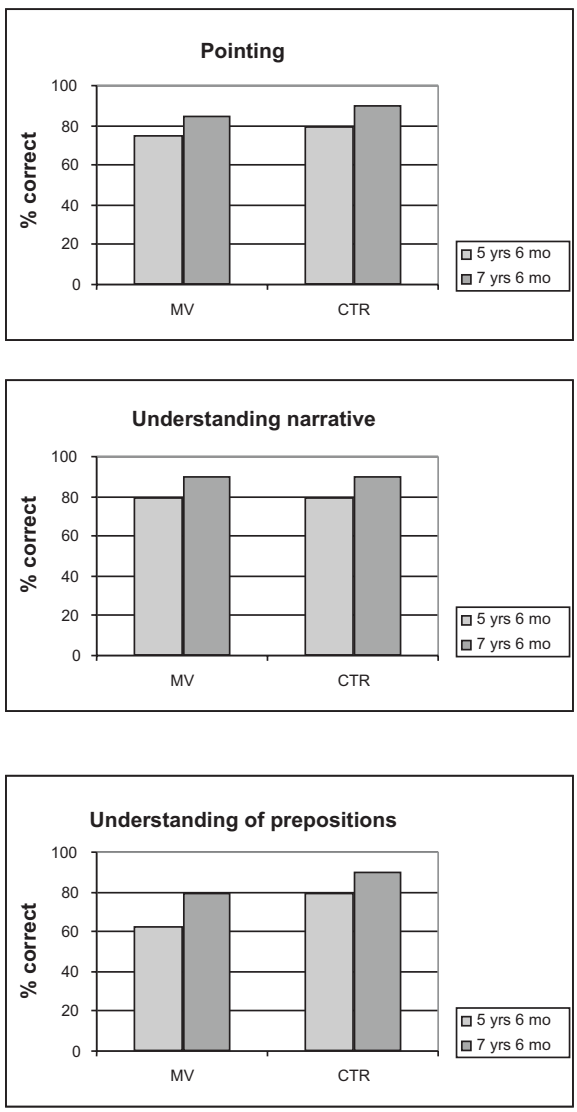

Fig. 2. Evolution of linguistic comprehension. Control norms are taken from Chevrie-Muller et al. (1981).

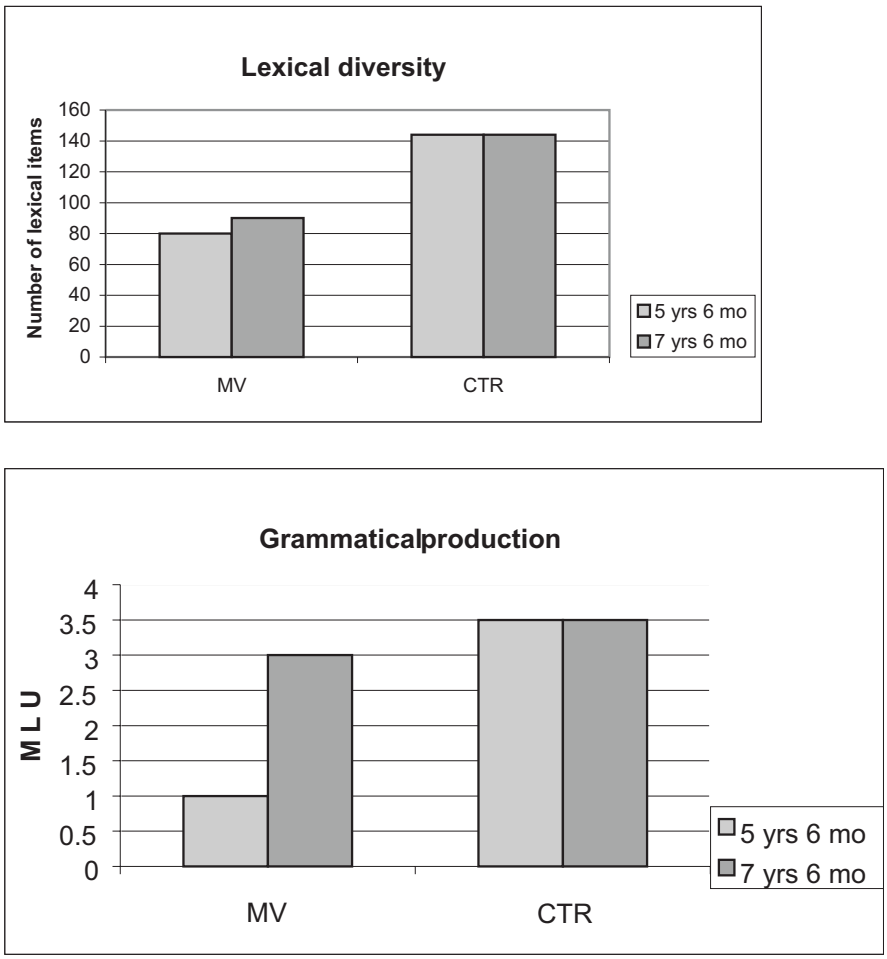

Fig. 3. Evolution of linguistic production. Control norms are taken from Le Normand (1997).

cerebral damage. In the present case, it appears that there is a dissociation between comprehension performance and production abilities. To the extent that speech production depends upon motor capabilities, it appears that MV's neuromotor deficits are correlated with her steady lag or deficit in productive aspects of language. In contrast, comprehension abilities improve with age and, at 7 years and 6 months, are within normal range, suggesting that neuronal mechanisms may differentially mediate linguistic comprehension and production. In the present case study, neuromotor stagnation accompanies a severe linguistic production lag or deficit. Thus the question as to whether motor and language development follow a parallel course gets a partial answer: expressive aspects of language and speech may share common maturational mechanisms; receptive aspects of language may, to a certain extent, evolve independently.

The role of the corpus callosum in the development of fine-tuned motor control has been well established (Ramaeckers, 1991). The lack of transcallosal information, as is the case with unmyelinated callosal fibers or immature callosal structure, is often considered responsible for the slow bimanual coordination and poorly imitated movements often seen in cases of abnormal morphogenesis of the corpus callosum. The behavioral motor observations and the neurolog-

Table 1. MV's performance on picture names repetition test

Phonotactic structure	Picture name	English translation	Production at 5 years 6 months	Production at 7 years 6 months	Target word
Monosyllabic words					
(n = 10)	nez	nose	[ne]+	[ne]+	[ne]
CV	roue	wheel	[u]	[ru]+	[ru]
CV	clé	key	[e]	[kle]+	[kle]
CCV	glace	mirror	[ta]	[glas]+	[glas]
CCVC	gomme	rubber	[dɔ]	[kɔm]	[gɔm]
CVC	doigt	finger	[a]	[gwa]	[dwa]
CcV	table	table	[a]	[ta]	[tabl]
CVCC	arbre	tree	[a]	[nab]	[arbr]
VCCCV	chaise	chair	[tɛ]	[sɛ]	[ʃɛz]
CVC	train	train	[]	[r]	[trɛ̃]
CCV					
Bisyllabic words					
(n = 18)	bébé	baby	[bebe]+	[bebe]+	[bebe]
CVCV	lapin	rabbit	[a)]	[lap)]	[lapɛ̃]
CVCV	poupée	doll	[pupe]+	[pupe]+	[pupe]
CVCV	couteau	knife	[uo]	[kuto]+	[kuto]
CVCV	maison	house	[ɛ]	[mɛz]	[mɛzɔ̃]
CVCV	jambon	ham	[t]	[zb]	[žãbɔ̃]
CVCV	ciseaux	scissors	[tio]	[tio]	[sizo]
CVCV	bouchon	cork	[bu]	[bus]	[buʃɔ̃]
CVCV	bougie	candle	[bui]	[buzi]	[byži]
CVCV	piano	piano	[pano]	[pjano]+	[pjano]
CcVCV	voiture	car	[ay]	[watyr]	[vwatyr]
CcVCVC	casserole	pan	[aɔ]	[kasɔl]	[kasrɔl]
CVCCVC	banane	banana	[bana]	[bana]	[banan]
CVCVC	avion	aeroplane	[aj]	[aj]	[ɑvjɔ̃]
VCcV	brouette	wheelbarrow	[bue]	[buɛt]	[bruɛt]
CCVVC	fromage	cheese	[ama]	[frɔmaʃ]	[frɔmɑž]
CCVCVC	crayon	pencil	[a]	[krɛʃ]	[krɛjɔ̃]
CCVCV	stylo	pen	[io]	[tilo]	[stilo]
CCVCV					
Multisyllabic words	bouquet de fleurs	bouquet	[titloer]	[bukɛfoe]	[bukɛdfloer]
(n = 5)	parapluie	umbrella	[papi]	[paapi]	[paraplwi]
CVCVCCCV	sausisson	sausage	[sio]	[sisis]	[sosisɔ̃]
CVCVCCCV	cigarette	cigarette	[gaɛ]	[sigaɛt]	[sigarɛt]
CVCVCV	allumette	matches	[amɛ]	[aymɛ]	[alymɛt]
CVCVCVC					
VCVCVC					
	Percentage correct:		9% (3)	30% (10)	
	Percentage incorrect:		91% (30)	70% (23)	

ical evaluation of patient MV appear to represent a good illustration of the interaction between structural deficit to the corpus callosum and functional motor deficits. Dennis (1981) has also shown that the integrity of the corpus callosum may be essential for the normal development of specific domains within language (e.g., syntax).

Although a diagnosis of severe speech production disorder can be made, a similarity is seen with the regular process of acquisition of speech sounds, when the articulatory aspect of MV's consonantal stages are considered. For example stops (e.g., /b, p/) were acquired before fricatives (/v,f/), and labials (/m,n/) before linguals (/l,r/) as is shown in Table 1. At any

rate, the present case is the first reported with a near total absence of consonants in speech.

In the case of MV, it is important to note that oropharyngeal motor actions in all other functions, besides consonant usage, were normal. Thus, considering that MV had acquired the syllabic structures, used meaningful combinatorial speech, and had a normal comprehension of words and sentences suggests that her problem is not one of perception and discrimination difficulties with speech sounds. A plausible explanation for her deviant linguistic development may lie in an impaired ability to plan and organize the coordinated sequences of the oropharyngeal movements that are necessary for complex speech production. The longer the words, the more difficult for MV to utter them correctly.

In conclusion, the present follow-up study shows that specific functions within the language domain are asynchronous in their acquisition and development. With the case of MV, a child with developmental dyspraxia, it is clear that production aspects of language were severely impaired and followed a parallel course with her neuromotor development. In contrast, receptive aspects of language, apparently independent of motor input, followed a normal sequence in her development. This case study suggests that motor development and motor control of speech appear to be under the control of the same maturational constraints.

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