INT J LANG COMMUN DISORD, XXX 2014,
VOL. 00, NO. 0, 1–17

Research Report

Spoken language comprehension of phrases, simple and compound-active sentences in non-speaking children with severe cerebral palsy

Joke J. M. Geytenbeek†§||, Margriet J. M. Heim‡, Dirk L. Knol‡, R. Jeroen Vermeulen¶|| and Kim J. Oostrom||#

†Department of Rehabilitation Medicine, VU University Medical Center, Amsterdam, the Netherlands
‡University of Amsterdam, Amsterdam, the Netherlands
§The EMGO Institute for Health and Care Research, VU University Medical Center, Amsterdam, the Netherlands
¶Department of Paediatric Neurology, VU University Medical Center, Amsterdam, the Netherlands
||Neuroscience Campus Amsterdam, Amsterdam, the Netherlands
#Department of Pediatric Psychology, VU University Medical Center, Amsterdam, the Netherlands

(*Received April 2014; accepted November 2014*)

Abstract

Background: Children with severe cerebral palsy (CP) (i.e. 'non-speaking children with severely limited mobility') are restricted in many domains that are important to the acquisition of language.
Aims: To investigate comprehension of spoken language on sentence type level in non-speaking children with severe CP.
Methods & Procedures: From an original sample of 87 non-speaking children with severe CP, 68 passed the pre-test (i.e. they matched at least five spoken words to the corresponding objects) of a specifically developed computer-based instrument for low motor language testing (C-BiLLT), admitting them to the actual C-BiLLT computer test. As a result, the present study included 68 children with severe CP (35 boys, 33 girls; mean age 6;11 years, SD 3;0 years; age range 1;9–11;11 years) who were investigated with the C-BiLLT for comprehension of different sentence types: phrases, simple active sentences (with one or two arguments) and compound sentences. The C-BiLLT provides norm data of typically developing (TD) children (1;6–6;6 years). Binomial logistic regression analyses were used to compare the percentage correct of each sentence type in children with severe CP with that in TD children (subdivided into age groups) and to compare percentage correct within the CP subtypes.
Outcomes & Results: Sentence comprehension in non-speaking children with severe CP followed the developmental trajectory of TD children, but at a much slower rate; nevertheless, they were still developing up to at least age 12 years. Delays in sentence type comprehension increased with sentence complexity and showed a large variability between individual children and between subtypes of CP. Comprehension of simple and syntactically more complex sentences were significantly better in children with dyskinetic CP than in children with spastic CP. Of the children with dyskinetic CP, 10–13% showed comprehension of simple and compound sentences within the percentage correct of TD children, as opposed to none of the children with spastic CP.
Conclusion & Implications: In non-speaking children with severe CP sentence comprehension is delayed rather than deviant. Results indicate the importance of following comprehension skills across all age groups, even beyond age 12 years. Moreover, the subtype of CP should be considered when establishing an educational programme for sentence comprehension, and augmentative and alternative communication support. In addition, educational programmes for children with severe CP should take into account the linguistic hierarchy of sentence comprehension when focusing on the input and understanding of spoken language comprehension.

Keywords: language development, cerebral palsy, spoken language comprehension, sentence comprehension, non-speaking.

Address correspondence to: Joke Geytenbeek, VU University Medical Center, Department of Rehabilitation Medicine, Reception B, PO Box 7057, NL-1007 MB Amsterdam, the Netherlands; e-mail: J.geytenbeek@vumc.nl

International Journal of Language & Communication Disorders
ISSN 1368-2822 print/ISSN 1460-6984 online © 2015 Royal College of Speech and Language Therapists
DOI: 10.1111/1460-6984.12151

What this paper adds?

What is already known on this subject?

Children with severe CP (i.e. non-speaking children with severely limited mobility) are restricted in many domains involved in the acquisition of language. Although studies on global measures of receptive grammar show that scores of children with severe CP are within the average range for their chronological age, many of these children still show extensive deficits in language comprehension.

What this paper adds?

This is the first in-depth study of sentence comprehension in non-speaking children with severe CP using a specifically developed instrument for the assessment of comprehension of spoken language. For children with severe CP who have reached a basic linguistic level (matching at least five spoken words to the corresponding objects), sentence comprehension appears to develop up to at least age 12 years and tends to follow the developmental trajectory of TD children, albeit at a much slower rate. Sentence types with additional elements (e.g. spatial relations and function words embedded in simple sentences) and compound sentences showed not only more discrepancy compared with TD children, but also significant differences in performance between individual children and between the subtypes of CP. Of the children with dyskinetic CP who showed better comprehension of syntactically more complex sentences, 10–13% achieved scores within the average range of TD children. In contrast, all children with spastic CP showed poor comprehension of the more complex sentences.

Introduction

Cerebral palsy (CP) is a permanent non-progressive disorder of the development of movement and posture caused by damage to, or malformation of, the developing brain in early life (Rosenbaum *et al.* 2007). With a prevalence of approximately 2.0 in every 1000 live born children in Europe, CP is the most common cause of physical disability in children (Andersen *et al.* 2008). Brain lesions in children with CP are characterized by their pathogenic events at different times during early brain development. The impact of these lesions on brain function can result in different functional outcomes in CP (such as type and severity of the physical disability) and in associated impairments (i.e. language and/or cognitive impairment) (Krageloh-Mann *et al.* 2002, Rosenbaum *et al.* 2007). CP can be classified according to the type of underlying gross motor movement impairment, i.e. spastic, dyskinetic and ataxic (Gainsborough *et al.* 2008). In addition, systems are available to classify the functional abilities of children with CP based on their self-initiated movement with an emphasis on walking, sitting, transfers and mobility (Gross Motor Classification System—GMFCS) (Palisano *et al.* 2000) and on how children use their hands to handle objects in daily activities (Manual Ability Classification System—MACS) (Eliasson *et al.* 2006). The more assistance or adaptation required, the higher the GMFCS and/or MACS level (see appendix A for details).

CP is a complex disorder, showing a large variety in children's expressive and receptive communication skills. Unique for CP is that motor impairments may explain motor speech problems (i.e. dysarthria or anarthria), but motor speech problems or absence of speech do not account for intellectual disabilities. On the other hand, a majority of children with CP exhibit associated impairments, such as intellectual disabilities that, apart from the motor speech problems, may affect language development and communication abilities. Although a definitive connection between speech motor involvement, cognitive/language involvement and gross motor involvement has not been established (Hustadt *et al.* 2014) a relationship between GMFCS level and associated impairments, such as communication difficulties and cognitive impairment, has been demonstrated (Andersen *et al.* 2008, Parkes *et al.* 2010, Sigurdardottir and Vik 2008, Sigurdardottir *et al.* 2011, Voorman *et al.* 2010). Higher GMFCS levels (i.e. level IV or V) are associated with more restrictions in cognition and communication (Voorman *et al.* 2010, Vos *et al.* 2014, Van Schie *et al.* 2013). To what extent the communication difficulties are the result of the motor speech problems, on the one hand, and the cognitive/language impairments, on the other, remains unclear. As a consequence of the motor speech and/or cognitive impairments, children with severe CP are confronted with restrictions in daily communication, thereby challenging their language development (Drager *et al.* 2010, Light 1997).

Theories of language development emphasize the interrelation between speech and language development, and language and cognitive development (Clark 2004, Hoff 2009).

Although it is unknown how language interacts with cognition, a hallmark of modern cognitive science is the goal of developing a theory of cognition that is powerful enough to encompass all human mental abilities, including language abilities (Harris 2003). Two major directions in psycholinguistic theories are the 'innate' theory, addressing language ability separately from cognition, and the cognitive/constructive theory considering language as a form of cognition. Current theories appear to be best captured by the idea that cognition and language have complex similarities and differences, and both develop over the human life span from genetic factors constrained by environmental input and cultural factors (Harris 2003, Chater and Christiansen 2010).

Whereas for TD children the process of language acquisition evolves rapidly and seemingly 'easy', for children with severe CP this process is difficult and less obvious (Drager *et al.* 2010, Light 1997). In children with severe CP, speech and language problems arise from deficits in speech-motor control, cognition, sensation/perception or a combination of these.

For instance, the first and later occurrence of vocal and conversational exchanges with parents or caregivers (including their feedback on the appropriateness of spoken words) is less often experienced in children with severe CP (Light 1997). Communication partners of TD children determine their level of language input based on the language produced by the child (Light 1997, Light and McNaughton 2011), whereas for communication partners of non-speaking children with severe CP this level of language input cannot be based on the language production of their child. In this context, it is not surprising that high levels of parental directiveness and restricted conversation patterns with children with severe CP are observed (Heim 2001, Pennington and McConachie 1999).

Furthermore, children with severe CP are limited in exploring the world on their own (Falkman *et al.* 2002, Light 1997, Light and Kelford Smith 1993). Considering that children with severe CP are restricted in their independent access to the environment and to their experience with the physical elements of the world (such as books, toys, kitchen/cooking and writing materials, dress-up clothes, and playing on the ground), their experiential basis for conceptual and lexical development is poor (Light and Kelford Smith 1993). As a consequence, it becomes more difficult for these children to acquire new knowledge and concepts about the world around them (Light 1997, Light and McNaughton 2011). All these factors limit the possibility of interaction with the (social and educational) environment and may jeopardize the development of linguistic skills necessary to participate in more complex interactive communication.

However, the fact that it may be difficult for children with severe CP to show understanding of language does not necessarily imply that they cannot develop comprehension of spoken words (receptive vocabulary) and, later, sentences (receptive grammar) (Bishop *et al.* 1990, Geytenbeek *et al.* 2010b, Light 1997). For children with severe CP for whom expressive speech is not possible, comprehension of spoken language contributes to communicative exchanges and is essential for the development of the child's language and communication abilities (Sevcik 2006, Light 1997, Drager *et al.* 2010). Moreover, comprehension skills are fundamental to social and academic success (Light 1997) and offer a basis for acquiring meanings of graphic and other non-vocal symbols. Obviously, assessment of spoken language comprehension abilities in children with severe CP is important, also because this knowledge provides important information for the interaction with parents/caregivers, teachers, and other communication partners (Geytenbeek *et al.* 2010b, Light 1997). Moreover, the level of spoken language comprehension can have important implications for the development of the child's augmentative and alternative communication (AAC) system and/or an individual education programme (Light 1997), Geytenbeek *et al.* 2010b). For instance, the use of AAC can enhance daily communication in non-speaking children and endorse the acquisition of meanings of spatial verbs, prepositions, particles and case markings they hear from their parents and/or caretakers (Drager *et al.* 2010). Studies on early receptive spoken language acquisition in non-speaking children with CP are scarce and the results are conflicting. Some authors using common measures of receptive grammar report age-adequate performance in severe CP (Andersen *et al.* 2008, Bruno and Trembath 2006), whereas others claim extensive delays and deficits in language comprehension (Bishop *et al.* 1990, Pirila *et al.* 2007). These inconsistencies may, at least in part, be explained by heterogeneity of the study sample (e.g. in motor type and/or severity of CP, or in age) or by methodological issues such as the tests used or the selection of outcome measures.

It is difficult to assess children with severe CP reliably because of the motor requirements associated with cognition and language testing (Geytenbeek *et al.* 2010a, 2010b, Yin *et al.* 2013). Indeed the development of a reliable assessment tool for broader level of cognitive functioning in children with severe CP is preferred; however, it is reported that measures of language comprehension can provide a distinct estimation of skills that is less influenced by the motor impairment of the child with severely limited mobility than nonverbal cognition measures (Ross and Cress 2006, Sigurdardottir *et al.* 2011).

On the other hand, commonly used standard language tests for comprehension of spoken language often need adaptations for the assessment of (young) non-speaking children with severe CP (Geytenbeek *et al.* 2010a, Pirila *et al.* 2007, Hustadt *et al.* 2014). Permitted adjustments are often minimal and difficult to establish in the administration of tests to children with severely limited mobility. In addition, non-standardized adjustments of the administration of tests can limit the generalizability and validity of comparisons of groups to a standard (Geytenbeek *et al.* 2010).

To address these issues, in the present study, the specifically designed Computer-Based Instrument for Low Motor Language Testing (C-BiLLT) was used. It requires minimal, if any, motor action and is a psychometrically sound instrument for the assessment of spoken language comprehension in non-speaking children with severe CP (for details, see Geytenbeek *et al.* 2014). The present study focuses on non-speaking children with severe CP, defined as children with functional limitations classified as GMFCS levels IV and V. The present study is part of ongoing research on the development and implementation of the C-BiLLT, and the investigation of spoken language comprehension in non-speaking children with severe CP, in relation to the underlying motor type of CP (i.e. spastic or dyskinetic), MRI findings, and related disabilities. The aim of the present study is to conduct an in-depth linguistic analysis of spoken sentence comprehension pertaining to phrases and simple and compound active sentence types in non-speaking children with severe CP compared with children with typical development.

Methods

Participants

An original sample of 87 nonspeaking children with severe CP was recruited from rehabilitation centres, special schools and special daycare centres for the physically handicapped throughout the Netherlands. The pre-test of the C-BiLLT is used to explore whether a child is able to communicate a choice between two concrete objects (or digital photographs of the objects) when provided with the spoken name of the objects. If the child is able to identify at least five out of eight objects or photographs correctly, the child proceeds to the computer test part of the C-BiLLT (see appendix B for a detailed description of the C-BiLLT). Of the 87 children, 19 (22%) children (nine boys, 10 girls; mean age 6;1 years, SD 2;6 years; age range 1;11–11;1 years) did not pass the pre-test of the C-BiLLT whereas 68 did, admitting this latter group to the actual C-BiLLT computer test. As a result, the present study included 68 children with severe CP (35 boys, 33 girls; mean age 6;11 years, SD 3;0 years; age range 1;9–11;11 years). Table 1 presents the demographic and medical characteristics of these children.

Inclusion criteria were: diagnosed with CP, GMFCS level IV or V, chronological age between 1;6 and 12;0 years, and anarthria (productive vocabulary fewer than five words). Children with severe visual impairment (visual acuity less than –3 of the better eye, diagnosed cerebral visual impairment or blindness) and/or hearing impairment (less than 30 dB in the better ear), and children without Dutch-speaking parent(s) were excluded.

The Medical Ethics Committee of the VU University Medical Centre Amsterdam approved the study. All parents provided written informed consent.

Spoken sentence comprehension

The present study evaluates seven sentence types (categorized in broader categories of 'linguistic mapping' and 'active sentence types pertaining to complex syntactic analyses'). Of the total 75 C-BiLLT computer items, 60 were used in the present study as they pertained to the sentence types of interest. Moreover, the remaining 15 items pertain to other sentence types and, because the item count per type category was fewer than five for these 15 remaining items, they were left out of consideration due to methodological issues. More specifically, we concluded that a statement about comprehension of a specific sentence type category in children with severe CP was only justified when this specific sentence type contained at least five items or more.

The seven sentence types under study pertain to the following:

Linguistic mapping:

- 'Noun phrases' (NP) that refer to animals, objects or persons, e.g. (in Dutch) '*waar is de auto?*' (where is the car?), '*waar is de baby?*' (where is the baby?) (20 items).
- 'Verb phrases' (VP) that refer to identification of infinitive verbs, e.g. '*wie is er aan het slapen?*] (Who is sleeping?) (10 items).

Active sentence types pertaining to complex syntactic analyses:

- Identification of persons presented in 'who' questions (WhQ) in combination with function of activity (e.g. '*wie maakt er muziek?*' (Who is making music?) (five items).
- Spatial relations embedded in simple sentences (SR) (e.g. '*de hond ligt in de mand*' ('the dog lies *in* the basket) (six items).
- Identification of simple sentences referring to non-observable situations (SOOC) of four persons (mother, baby, son Joshua and daughter Kim) (e.g. '*Joost en Kim gaan buitenspelen, wie blijft er bij mama?*' (Joshua and Kim are going to play outside, who stays with Mammy?) (six items).
- Identification of simple sentences with function words (SSF) (e.g., articles, prepositions, adjectives), quantifiers and comprehension of two or more concepts in one sentence referring to food products (e.g. '*Een kleine pot jam staat naast de*

Table 1. Characteristics of the children with severe cerebral palsy and statistical differences in means or proportions between children with spastic and dyskinetic CP

Variables	*n* (%)	Spastic (%)	Dyskinetic (%)	*p*
Number of children	68	31(46)	37 (54)	
Mean age (years)/SD	6;11/3;0	7;6/2;11	6;5/3;2	0.970
Age range (years;months)	1;7–12;0	2;2–12;0	1;7–12;0	0.158
Gender				0.611
Male	35 (51)	17(55)	18(49)	
Female	33 (49)	14(45)	19(51)	
GMFCS level				0.701
IV	29 (43)	14(45)	15(41)	
V	39 (57)	17(55)	22(59)	
MACS level				0.042*
< 4 years	15 (22)	4(13)	11(30)	
II	3 (4)	2 (6)	1 (3)	
III	13 (19)	10(32)	3 (8)	
IV	14 (21)	4(13)	10(27)	
V	23 (34)	11(36)	12(32)	
Gestational age				0.000***
< 37 weeks	18 (26)	15(48)	3 (8)	
≥ 37 weeks	50 (74)	16(52)	34(92)	
Range	24–42	24–42	26–42	
Parental level of education				0.538
Low	6 (9)	4(13)	2 (5)	
Middle	11 (16)	4(13)	7(19)	
High	43 (63)	20(64)	23(62)	
Missing	8 (12)	3(10)	5(14)	
Epilepsy				0.045*
Present	20 (29)	13(42)	7(19)	
Absent	47 (69)	18(58)	29(78)	
Missing	1 (2)	–	1 (3)	

Notes: CP = cerebral palsy; SD = standard deviation; GMFCS = gross motor function classification system; MACS = manual ability classification system.
$^{*}p < 0.05$, $^{**}p < 0.01$, $^{***}p < 0.001$.

rode pot jam' (A little jar of jam is standing beside the red jar of jam) (seven items).

- Identification of compound sentences (CS) referring to the lunch table (e.g. '*Eerst lag er een appel op tafel maar nu is er een banaan voor in de plaats gekomen*' (First an apple was lying on the table, but now there is a banana in its place) (six items).

An overview of all 60 items are available on request from the first author (see appendix C for examples of visual presentation of the different sentence types).

Procedure

The C-BiLLT computer test administration took place in a distraction-free room at the child's rehabilitation centre, special school, special daycare centre or at the outpatient clinic of the Department of Rehabilitation Medicine, VU University Medical Center, Amsterdam. An experienced speech pathologist (JG) performed the C-BiLLT administrations in the children with severe CP. Although the speech pathologist was allowed to stimulate and encourage the child in any way possible, the actual question or sentence could only be repeated once. Each question or sentence was spoken with as near natural prosody as possible. The response of the children was scored correct or false: a response was noted correct if the child convincingly had activated an input switch and stopped pushing or touching it for more than 3 s, or the child had pointed to or touched the touch screen with his/her hand, foot, arm or elbow, or had been (eye) gazing at the image for more than 2 s. The switches were used by 22 (32%) children either with their own wheelchair head support ($n = 16$) or with the switches touched by any body part ($n = 6$). Direct selection was used by 40 (59%) children either by the touch screen function ($n = 26$, 38%) or with eye-gazing ($n = 14$, 21%). Six children (9%) used a combination of direct selection and switches. The test was stopped after eight successive incorrect responses or if the child was no longer cooperative or no longer made any visual contact with the (touch) screen or had been clearly inattentive. On average the C-BiLLT assessment took 15–60 min. For each correctly answered item one point is scored with a maximum of 60 points.

Different outcome scores were calculated: (1) seven individual sum scores, i.e. a sum score per sentence type (NP, VP, WhQ, SR, SOOC, SSF and CS); (2) a 60-item total sentence comprehension score; and (3) percentage correct scores per sentence type (100 × [number of correct items pertaining to that specific sentence type/ total items pertaining to that specific sentence type]).

Associated impairments, risk and social factors

Because the impact of brain lesions on brain function can result in different outcomes and associated impairments (Krageloh-Mann *et al.* 2002), type of CP, gestational age (GA) and epilepsy were independent factors to be investigated. GA was categorized into preterm (GA < 37 weeks) and term born (GA ≥ 37 weeks). Type of CP was defined according to the guidelines of the Surveillance of Cerebral Palsy in Europe (SCPE) (Gainsborough *et al.* 2008). Epilepsy was defined as diagnosed epilepsy after the neonatal period of the child and reported accordingly in medical reports. Additionally, with respect to language development, studies have shown that girls develop faster than boys, and that children from families with a higher level of education generally develop faster than children from lower level of education (Wassenberg *et al.* 2008, Zambrani *et al.* 2012). Therefore, differences between sentence comprehension in boys and girls, and the possible influence of parental level of education, were also investigated. Parental level of education was defined as low (practical education and lower), medium (theoretical education and upper secondary vocational education) and high (secondary non-vocational, higher education and university).

Statistical analyses

Descriptive statistics were used to describe the main characteristics of the children with severe CP. Per CP subgroup, Fisher's exact tests were used to analyse differences in proportions of GMFCS and MACS level (the latter only for children aged more than 4 years), GA, epilepsy, gender and parental levels of education.

Sentence type comprehension in children with CP versus TD children

Spearman rank (rho) was used to investigate the correlation between chronological age and the 60-item total sentence comprehension score.

A binomial logistic regression was performed to reveal trends in sentence type comprehension (corrected for over-dispersion) between children with CP (reference group) and TD children. Sentence type percentage correct scores were dependent variables and covariates were group (CP or TD), chronological age and chronological age (squared), and interactions group × chronological age and group × chronological age (squared). Analyses were based on the assumption that every item within a certain sentence type has equal difficulty.

Table 2. Age distribution of the total sample of children

	Children with CP			TD children
Age (years)[a]	*n*	(age band)[b]	*n*	*n*
1	2			45
2	5	(1–3)	15	129
3	8			137
4	6			191
5	4	(4–6)	23	202
6	13			91
7	4			11
8	5	(7–9)	14	–
9	5			–
10	8			–
11	8	(10–12)	16	–
12	0			–
Total	68			806

Notes: CP = cerebral palsy; TD = typical development.
[a]Chronological age.
[b]Age band per 3 years.

Sentence type comprehension within CP

To increase power and to improve the sampling distribution of the children with severe CP, regression analyses were also carried out in broadened age groups (grouped per age 'bands' of 3 years; table 2). A binomial logistic regression was used to analyse differences in comprehension of the different sentence types between different age bands and subtype of CP. Each sentence type percentage correct was the dependent variable and age band, type of CP and the interaction age band × type of CP were the covariates. Age band 4 (10–12 years) and dyskinetic CP were used as the reference. Outcomes were denoted in odds ratios.

Because CP is characterized by its variability in clinical outcomes between children, we also report on individual differences in sentence type comprehension in children with severe CP. We agreed that comprehension of a sentence type is considered to be 'mastered' if, in the TD group, within a certain chronological age group at least 85% of the items pertaining to a particular sentence type was answered correctly.

In addition, to investigate whether children with different gross motor function and manual ability skills would perform differently on comprehension of spatial relations, a binary logistic regression analysis was performed. The outcome variable was dichotomized in 'mastered' and 'not mastered'. Predictors were level of GMFCS (IV [as reference] and V) and MACS (levels II/III combined [as reference] and levels IV/V combined), type of CP (with spastic CP as reference) and chronological age.

To determine which independent variables (type of CP, GMFCS and MACS (II/III and IV/V combined, gender, GA, parental education and epilepsy) predict

Table 3. Binomial logistic regression of sentence types between children with severe CP and TD children, expressed in percentage correct per sentence type

	CP children			TD children		Difference CP versus TD
Sentenc type	Age[a]	P	95% CI	P	95% CI	*p*
NP	1	81	68–90	70	65–75	0.011
	2	81	72–87	94	92–96	< 0.001
	3	81	77–84	100	98–100	< 0.001
	4	81	76–85	100	98–100	< 0.001
	5	82	77–86	100	98–100	< 0.001
	6	84	78–88	100	98–100	< 0.001
	7	85	80–89	100	98–100	< 0.001
	8	91	83–91	–		
	9	90	85–93	–		
	10	92	86–95	–		
	11	93	87–97	–		
VP	1	65	51–77	53	49– 5	0.041
	2	72	64–79	88	85– 90	< 0.001
	3	78	73–83	99	98– 100	< 0.001
	4	83	78–86	100	98– 100	< 0.001
	5	86	81–89	100	98– 100	< 0.001
	6	88	83–91	100	98–100	< 0.001
	7	89	86–92	100	98–100	< 0.001
	8	90	87–93	–		
	9	91	87–93	–		
	10	90	86–93	–		
	11	89	83–94	–		
WhQ	1	21	11–38	21	18–25	0.913
	2	29	19–41	39	35–42	0.115
	3	36	29–45	58	55–60	< 0.001
	4	44	37–51	72	70–75	< 0.001
	5	51	43–59	82	80–84	< 0.001
	6	57	49–65	88	86–89	< 0.001
	7	62	54–69	91	90–92	< 0.001
	8	66	59–73	–		
	9	69	62–76	–		
	10	72	63–79	–		
	11	73	61–83	–		
SR	1	09	04–19	12	10–15	0.506
	2	13	08–19	23	21–26	0.050
	3	19	13–26	37	35–40	< 0.001
	4	26	20–32	52	50–55	< 0.001
	5	33	27–40	65	63–67	< 0.001
	6	40	33–47	75	73–76	< 0.001
	7	46	38–53	81	79–82	< 0.001
	8	51	44–58	–		
	9	55	49–62	–		
	10	59	51–66	–		
	11	61	49–71	–		
SOOC	1	04	00–18	04	03–06	0.749
	2	05	01–16	09	07–13	0.209
	3	08	04–14	18	15–21	0.004
	4	12	08–16	29	27–32	< 0.001
	5	15	11–20	43	39–45	< 0.001
	6	20	15–27	55	52–58	< 0.001
	7	26	19–33	66	63–68	< 0.001
	8	31	25–39	–		
	9	37	30–44	–		
	10	42	35–50	–		
	11	48	37–59	–		
SSF	1	n.a.	n.a.	03	02–04	n.a.

Continued

Table 3. ***Continued***

Sentenc type		CP children		TD children		Difference CP versus TD
	Age[a]	P	95% CI	P	95% CI	*p*
	2	n.a.	n.a.	08	06–09	n.a.
	3	n.a.	n.a.	15	13–17	n.a.
	4	08	05–10	27	25–29	< 0.001
	5	13	09–18	39	37–41	< 0.001
	6	20	15–27	51	49–53	< 0.001
	7	27	22–35	61	59–63	< 0.001
	8	34	27–41	–		
	9	39	32–46	–		
	10	41	34–49	–		
	11	42	32–53	–		
CS	1	n.a.	n.a.	03	02–04	n.a.
	2	n.a.	n.a.	06	05–08	n.a.
	3	n.a.	n.a.	11	09–13	n.a.
	4	08	04–12	18	17–20	< 0.001
	5	11	07–15	27	26–29	< 0.001
	6	15	11–22	37	35–39	< 0.001
	7	20	15–28	45	43–48	< 0.001
	8	26	20–38	–		
	9	32	26–38	–		
	10	37	30–45	–		
	11	42	31–54	–		

Notes: [a]= Chronological age (years); P = percentage correct; CP = cerebral palsy; TD = typical developing children; CI = 95% confidence interval; NP = noun phrase; VP = verb phrase; WhQ, 'who' questions; SR = spatial relations; SOOC = simple sentence out of context; SSF = simple sentence with function words; CS = compound sentence.
Model: intercept + group + age + age (squared) + group × age + group × age (squared); n.a. = not applicable.

the variability in total sentence comprehension score, a regression analysis (backward method) was conducted. Only variables that were significant in the univariate analyses or with $p < 0.1$ were used in the models. The models were made without interaction between GA × type of CP and epilepsy × type of CP because both variables were highly associated. Normal plots for residuals were made to check whether the residuals were approximately normally distributed. In all tests, the statistical significance level was taken as $p < 0.05$. All statistical analyses were performed with SPSS-20 for IBM (SPSS Inc. Chicago, IL, USA).

Results

Main characteristics of the children with CP

Table 1 presents the main characteristics of the children with CP. No significant difference in proportions were found for chronological age, gender, GMFCS or parental level of education. The distribution of MACS levels ($n = 53$) shows a significant difference between children with spastic and dyskinetic CP ($p = 0.042$), with higher MACS levels (IV and V) in children with dyskinetic CP. A significant association was found between MACS and GMFCS level ($p < 0.001$) with higher MACS levels (IV and V) in GMFCS V. The majority of the CP sample was born at term. A significant association was found between GA and type of CP ($p < 0.001$), with more children born preterm in spastic CP than in dyskinetic CP (15/31 versus 3/37). A significant association was found between epilepsy and type of CP ($p = 0.045$), with more epilepsy in children with spastic CP than in children with dyskinetic CP (13/31 versus 7/37).

Sentence type comprehension in children with CP versus TD children

Overall, the 60-item total sentence comprehension score in children with severe CP increased with age (rho = 0.49, $p < 0.001$) and tends to follow (albeit at a much slower rate) the developmental trajectory of TD children (rho = 0.82, $p < 0.001$).

The binomial logistic regression analyses (table 3) confirmed that chronological age modulated sentence type comprehension, both in TD children and in children with CP. TD children showed a steep increase in noun and verb phrases reaching ceiling levels of 100% correct at age 3 years. After a fixed percentage for the first 4 years, children with CP showed a gradual increase in comprehension of noun and verb phrases up to age 12 years, reaching percentages near the ceiling level. In children aged 1 year, the percentage correct was higher for children with CP ($n = 3$) than for TD children.

Table 4. Binomial logistic regression of percentage correct of sentence types between age band and subtype of CP ($n = 68$)

Sentence type	OR	CI	p
NP			
Age band 1 versus 4	0.297	0.060–1.475	0.137
Age band 2 versus 4	0.358	0.081–1.577	0.175
Age band 3 versus 4	0.648	0.115–3.664	0.624
Spastic versus dyskinetic	0.475	0.176–1.282	0.142
VP			
Age band 1 versus 4	0.403	0.104–1.555	0.187
Age band 2 versus 4	0.685	0.189–2.480	0.565
Age band 3 versus 4	2.23	0.345–14.395	0.400
Spastic versus dyskinetic	0.684	0.260–0.1.801	0.442
WhQ			
Age band 1 versus 4	0.143	0.044–0.470	< 0.001***
Age band 2 versus 4	0.444	0.161–1.221	0.116
Age band 3 versus 4	1.16	0.351–3.811	0.811
Spastic versus dyskinetic	0.410	0.185–0.911	0.029*
SR			
Age band 1 versus 4	0.060	0.016–0.2234	0.001*
Age band 2 versus 4	0.401	0.159–1.010	0.053
Age band 3 versus 4	0.656	0.236–1.826	0.420
Spastic versus dyskinetic	0.379	0.163–0.879	0.024*
SOOC			
Age band 1 versus 4	0.007	0.000–0.180	0.003**
Age band 2 versus 4	0.306	0.118–0.793	0.015*
Age band 3 versus 4	0.294	0.099–0.872	0.027*
Spastic versus dyskinetic	0.212	0.090–0.501	0.001***
SSF			
Age band 1 versus 4	–	–	–
Age band 2 versus 4	0.333	0.193–0.575	0.001***
Age band 3 versus 4	0.470	0.259–0.853	0.013*
Spastic versus dyskinetic	0.373	0.232–0.600	0.001***
CS			
Age band 1 versus 4	–	–	–
Age band 2 versus 4	0.253	0.106–0.607	0.002**
Age band 3 versus 4	0.325	0.124–0.850	0.022*
Spastic versus dyskinetic	0.161	0.071–0.367	0.001***

Notes: Age band 1 = child aged 1–3 years; Age band 2 = child aged 4–6 years; Age band 3 = child aged 7–9 years; Age band 4 = child aged 10–12 years; OR = odds ratio of percentage correct score; CP = cerebral palsy; CI = confidence interval; NP = noun phrase; VP = verb phrase; WhQ = 'who' questions; SR = spatial relations; SOOC = simple sentence out of context; SSF = simple sentence with function words; CS = compound sentence.
*$p < 0.05$, **$p < 0.01$, ***$p < 0.001$.

Comprehension of 'who' questions lagged behind in children with CP compared with TD children. TD children showed a steady increase in percentage correct up to age 6 years, reaching 88%, whereas children with CP showed increasing percentages correct up to 73% at age 11 years (a percentage almost reached at age 4 years in TD children). With an increase in sentence complexity, children with CP fell behind TD children and showed a decline in percentage correct. This decline in percentage correct was also observed in TD children; however, at age 7 years, TD children reached higher percentages for these complex sentence types than children with CP aged 11 years.

Sentence type comprehension within CP

When comparing types of CP, a significant difference in comprehension of sentence types was found for all sentence types pertaining to complex syntactic analyses, with better outcomes for children with dyskinetic CP (table 4 and figure 1). Significant differences were found between age band 1 and age band 4 for all complex syntactic sentence types, and between all age bands for simple sentences out of context (SOOC), simple sentence with function words (SSF) and for compound sentences (CS). For verb phrases and 'who' questions, age band 3 performed slightly better than age band 4 (difference not significant). As expected, sentence types SSF and CS were not applicable in children in age band 1.

On an individual level, children with dyskinetic CP mastered the different sentence types at a younger age than children with spastic CP; also, some children with dyskinetic CP (13% and 10%, respectively), observed from the age of 6;5 years, mastered the complex syntactic sentences in contrast to none of the children with spastic CP (table 5).

As revealed by the binary logistic regression analysis, neither MACS nor GMFCS classification had an effect on mastering the spatial relations sentences; the only significant predictor was the type of CP. The odds of a child mastering the spatial relation sentence type and having dyskinetic CP was 8.8 times higher than for a child with spastic CP ($p = 0.003$, OR = 8.826, CI = 1.64–62.81).

No significant differences in total sentence comprehension score were found between boys and girls ($p = 0.257$), MACS levels ($p = 0.797$), epilepsy ($p = 0.882$) and parental education ($p = 0.775$). In both subgroups of CP, parental level of education was most dominantly represented by the highest level of education (> 60%).

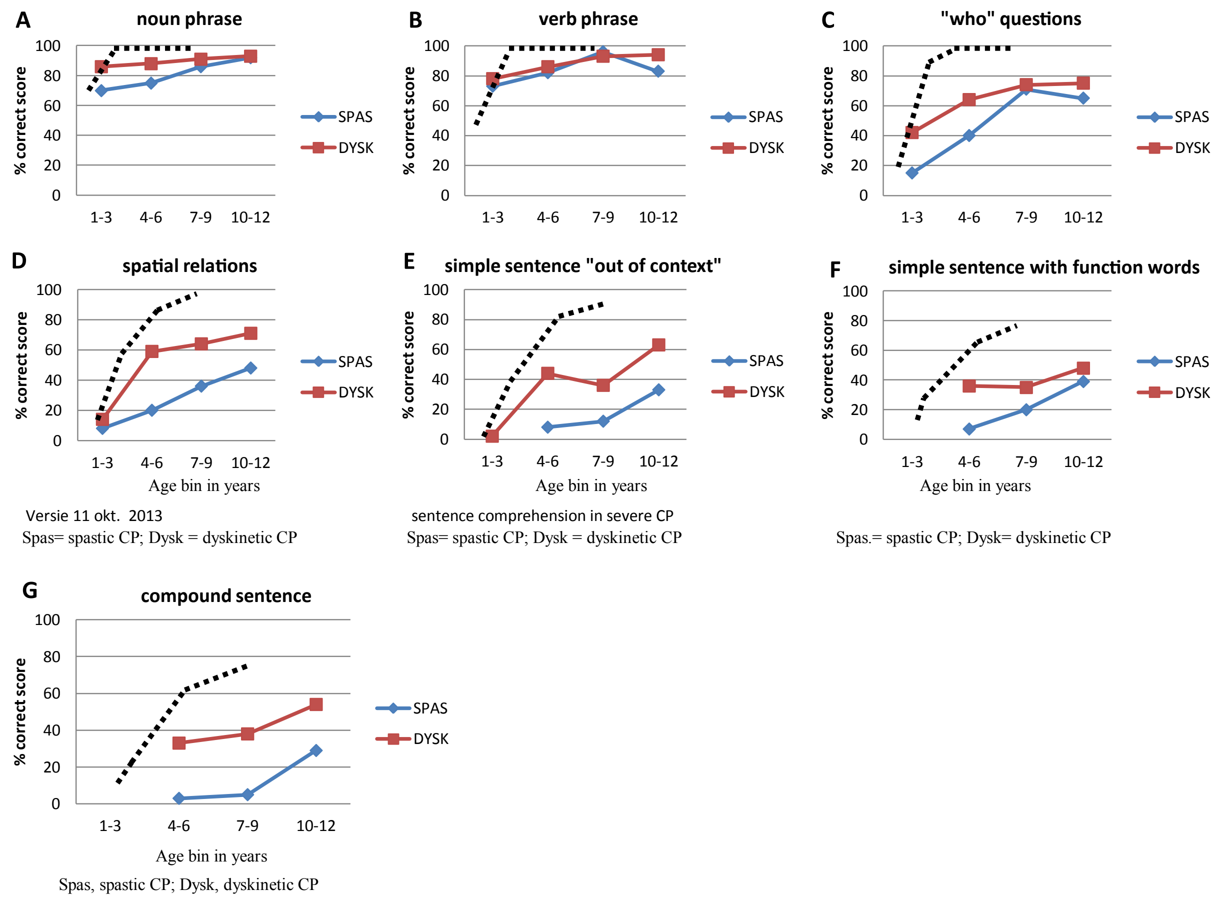
The regression analysis revealed that the chronological age of the child, type of CP and level of GMFCS explained 37% of the variance in total sentence comprehension score (table 6).

Discussion

This study investigated comprehension of spoken language on a sentence level in non-speaking children with severe CP using a specifically designed instrument that meets test requirements for non-speaking children with severely limited mobility (GMFCS IV/V). The performance of children with CP was compared with that in TD children.

Sentence type comprehension in children with CP versus TD children

As expected, sentence comprehension scores were significantly poorer in children with severe CP than in TD children. On the other hand, results confirm the idea that the fact that a child with severe CP who cannot speak, does not necessarily imply that he/she is not able to develop comprehension of spoken sentences



Abbreviations: TD = typical development; CP = cerebral palsy; NP= noun phrase; VP= verb phrase; WHO= "who"questions; SR= spatial relations embedded in simple sentences; SOOC= simple sentences out of context; SSF= simple sentences with function words ; CS= compound sentences.

Figure 1. [A-G.] Observed percentage correct per sentence type in children with severe CP (coloured lines) and TD children (dotted line).

(receptive grammar). Moreover, sentence type comprehension in children with severe CP seems to follow the same trajectory as in TD children, but at a much slower rate. On the other hand, in our study sample, some children with dyskinetic CP followed the developmental trajectory at the same pace as TD children.

Sentence type comprehension within CP

In severe CP, comprehension of simple syntax (noun/verb phrases) showed a similar development between types of CP, whereas complex syntactic analyses was generally better in children with dyskinetic CP than in children with spastic CP. Vasilyeva *et al.* (2008) stated that 'the acquisition of obligatory aspects of simple syntax may rely on common mechanisms that are available to all children. The acquisition of complex syntax however, might reflect individual differences in a child's ability to extract complex structures from input'. In view of the role of the environment and of parents/caregivers, basic aspects of simple syntax appear regularly in everyday speech while the acquisition of *complex syntax* most likely reflects the variability in linguistic environments that is available to the child. Therefore, the use of these structures and the amount/nature of the input may be critically related to the spoken language comprehension and cognitive abilities of an individual child. Interestingly, we found that the acquisition of *complex syntax* was also critically related to the type of CP, with higher comprehension scores for children with dyskinetic CP. This difference in complex sentence comprehension between the subtype of CP may partly be explained by the fact that, in children with severe CP, the spastic CP type appears to be associated with the severity of cognitive deficits while in children with dyskinetic CP this relation is less well established (Krageloh-Mann *et al.* 2002, Sigurdardottir *et al.* 2008). Indeed, it is thought that language input can influence cognitive development (Clark 2004). For instance, when a child is able to comprehend what is being labelled, then making similarities,

Table 5. Mean score and 85% correct score of individual children and CP groups of different sentence type, related to the subtypes of CP (31 spastic children, 37 dyskinetic children)

Sentence type (number of items)		Type of CP	Mean	(range)	SD	CI	Individual[a] ≥ 85% Age (years)	Group[b] ≥ 85% *n* (%)
NP	(20)	Spastic	16.29	(5–20)	5.29	14.35–18.23	3;2	21 (68)
		Dyskinetic	17.84	(6–20)	3.79	16.51–19.16	1;7	28 (76)
VP	(10)	Spastic	8.39	(0–10)	2.64	7.42–9.35	3;2	20 (65)
		Dyskinetic	8.65	(4–10)	2.10	7.95–9.20	3;2	25 (68)
WhQ	(5)	Spastic	2.42	(0–5)	1.73	1.79–3.05	6;3	3 (10)
		Dyskinetic	3.08	(0–5)	1.96	2.43–3.74	2;5	11 (32)
SR	(6)	Spastic	1.74	(0–6)	1.86	1.06–2.42	10;4	2 (6)
		Dyskinetic	2.95	(0–6)	2.32	2.17–3.72	4;4	14 (38)
SOOC	(6)	Spastic	0.87	(0–5)	1.43	0.35–1.40	11;0	1 (3)
		Dyskinetic	1.97	(0–6)	2.33	1.20–2.75	6;5	8 (22)
SSF	(7)	Spastic	1.23	(0–5)	1.86	0.54–1.91	Not Achieved	0 (0)
		Dyskinetic	1.95	(0–7)	2.35	1.16–2.73	6;5	5 (13)
CS	(6)	Spastic	0.58	(0–4)	1.15	0.16–1.00	Not Achieved	0 (0)
		Dyskinetic	1.73	(0–6)	2.05	1.05–2.41	6;5	4 (10)

Notes: Range = range in score; SD = standard deviation; CI = confidence interval; ind. = individual; NP = noun phrase; VP = verb phrase; WhQ = 'who' questions; SR = spatial relations; SOOC = simple sentence out of context; SSF = simple sentence with function words; CS = compound sentence.
[a]Youngest child observed achieving a score of 85% correct.
[b]Number of children within the CP group achieving a score of 85% correct.

Table 6. Multiple regression model of total sentence comprehension score of children with CP ($n = 68$)

	B	SE B	β
Chronological age (years)	0.199	0.039	0.52***
Spastic CP versus dyskinetic CP	9.476	2.809	0.34***
GMFCS IV versus V	−8.496	2.803	−0.30**

Notes: Dependent variable = total sentence comprehension score.
$R^2 = 0.37$, adjusted $R^2 = 0.34$.
**$p < 0.01$, ***$p < 0.001$.
CP = cerebral palsy; GMFCS = gross motor function classification system.

comparisons and conceptual relations (necessary to understand complex syntactical sentences) are more easily facilitated. It is also thought that cognition and language interact in a cyclic manner as children develop (Clark 2004, Harris 2003); from this viewpoint, one can expect a more favourable development in language comprehension in children without (severe) cognitive deficits.

Similarly, children with spastic CP had more difficulty in understanding spatial relations than children with dyskinetic CP. Strategies for organizing space seem well established in TD children aged 15–18 months (Clark 2004). When exploring the environment, TD children start their initial conceptual knowledge about spatial relations. This provides a starting point to learn about the meanings of spatial verbs, prepositions and particles that they hear from their caregivers when exploring their environment. Although physical restrictions for exploring the environment and organizing space were similar in children with spastic and dyskinetic CP, children with dyskinetic CP showed more understanding of spatial relations, suggesting a possible influence of cognitive skills. It is noteworthy, however, that understanding of spatial relations increased with increasing age in children with spastic and with dyskinetic CP, but most favourably in children with dyskinetic CP. Nevertheless, it seems that, despite severe motor impairments and cognitive deficits, some children with severe CP are able to detect conceptual representations of objects and relations.

Furthermore, of note is that GA was associated with the type of CP. The association between type of CP and GA may be attributed to the fact that, in preterm born children, white matter injury (periventricular leukomalacia—PVL) is the most common cause of CP leading to spastic CP, while in term born children, lesions of the basal ganglia and thalamus (grey matter injury) most often occur, leading to dyskinetic CP (Mercuri *et al.* 2002). As already mentioned, children with dyskinetic CP often have severe motor problems but less prominent cognitive deficits (Krageloh-Mann *et al.* 2002). On the other hand, severe PVL with bilateral extensive white matter loss leads to severe spastic CP and is related to severe cognitive impairment (Krageloh-Mann *et al.* 1999).

The periventricular white matter contains major intra-hemispheric connections between frontal and temporal language regions (Northam *et al.* 2012). This functional connectivity and maturation is necessary to fully establish a sophisticated language system, integrating lexical and syntactic competence (Perani *et al.* 2011). The brain lesions associated with PVL may, therefore, disrupt the integrity of this important language network and disturb further development of functional connectivity and maturation of intra-hemisperic fibre bundles (Volpe 2009). Brain lesions associated with central grey matter injury (i.e. basal ganglia) are originally more confined to motor planning and control. Recently, Barbas *et al.* (2013) reported a strong association of the basal ganglia with prefrontal as well as motor and pre-motor cortices, suggesting that control of movements (e.g. speech articulation) is more related to grey matter injury. This might explain why high rates of speech impairment are particularly associated with children with dyskinetic CP (Parkes *et al.* 2010), while verbal cognition may be relatively spared.

Indeed, a significant difference between the type of CP was found for all sentence types pertaining to complex syntactical analyses, with better performances in children with dyskinetic CP. This finding suggests that children with spastic CP experience more difficulty in integrating lexical, syntactical, morphological and prosodic cues necessary to understand more complex sentences.

Recent brain imaging studies identified more than 50 brain areas involved in various aspects of language and cognitive mechanisms (Binder *et al.* 2009, Price 2012, Straube *et al.* 2012). The proposed relation between language and cognition may be most relevant for the results emerging from the present study, suggesting a relation between the pathogenic event at different trimesters of gestation, the location and severity of lesions (causing different types of CP), and functional outcome in language comprehension on a sentence level. Sentence comprehension is a complex function that engages many components of the language network and other brain regions that support language, such as working memory and cognitive control systems (Friederici 2002, Dronkers *et al.* 2004). Studies examining the neurological basis for language comprehension indicate the involvement of an extensive network of cortical regions and white matter pathways (Turken and Dronkers 2011), which are likely to be more affected in children with spastic CP than in children with dyskinetic CP. Our group is currently examining MRI scans to more thoroughly investigate these relations in children with severe CP.

Associated impairments and social factors

The results of the regression analysis show that neither gender, nor the presence of epilepsy, nor manual (dis)ability contributed to the variability in sentence comprehension in children with severe CP. The contradiction between the finding that the GMFCS level contributed to the variability in total sentence comprehension, but not to the comprehension of spatial relations, might be explained by the finding that only children with dyskinetic CP mastered spatial relations, thereby overruling the influence of the severity of the motor impairment.

Furthermore, parental level of education did not appear to be of particular importance. Previous findings on complex sentence comprehension suggest that simple and more complex syntactic skills are related in different ways to educational environments (Vasilyeva *et al.* 2008). It seems that despite restrictions in production of speech or variability in language input (e.g. limited spoken language input or adult addressed language), non-speaking children with severe CP can learn and recognize a substantial amount of spoken vocabulary (Andersen *et al.* 2008, Bishop *et al.* 1990, Light 1997). On the other hand, the present study demonstrates that this development is severely delayed and shows significant differences between subtypes of CP and between individual children with severe CP.

The most convincing results are that the acquisition of sentence comprehension continued to develop up to at least age 12 years and that children with dyskinetic CP (as a group) performed considerably better than children with spastic CP. This finding is important and is in line with a growing body of literature on cognitive functioning in children with different types of CP.

Limitations of the present study

The following study limitations should be taken into account. First, although the TD sample for normed data was relatively extensive, ceiling effects may have occurred. We included TD children up to age 7 years and, as expected, found 100% correct scores for noun and verb phrases from age 3 years. Suboptimal scores were found in TD children for the more complex sentences. Because, group wise, no 100% correct scores were found in the CP sample, it is unlikely that ceiling effects obscured the findings in the CP sample. However, individually, both TD as well as (dyskinetic) children with CP did reach 100% correct scores for the more complex sentences. This emphasizes the importance of broadening the age range in the TD sample (as is done in our ongoing research). Second, because our group of children with CP was relatively small and with an uneven age distribution, the present results cannot be generalized and should be interpreted with caution. Third, in the present study, only the level of parental education was taken into account, and the quality and diversity of parental spoken language input was not investigated. Finally, levels of education were missing for 12% of the parents and relatively few parents had a low level of education. A more in-depth study of parental multimodal language input is clearly required to investigate whether language input and parental education have an effect on spoken sentence comprehension in children with severe CP.

Conclusions

The findings of the present study indicate that, as expected, sentence comprehension in children with severe CP is significantly poorer compared with TD children. Nevertheless, our data show that for those children with severe CP who passed the pre-test, the development of language comprehension continues up to at least age 12 years. Especially the comprehension of syntactically more complex sentences is delayed in children with severe CP. When comparing sentence comprehension within CP, a significant difference in performance was observed between individual children and between children with spastic and dyskinetic CP. The differences between individual children, but in particular between types of CP, demonstrate that it is essential to follow both a child's individual language performance across all age bounds (even beyond age 12 years) as well as to consider which type of CP the child has. Moreover, educational programs for children with severe CP should consider the linguistic hierarchy of sentence comprehension when focusing on the development and understanding of spoken language comprehension.

Early detection of comprehension skills and adequate support for parents and/or caregivers of the child with severe CP can have important implications regarding how to address the child and augment the language comprehension performance of the child. Moreover, with early and adequate knowledge of a child's comprehension abilities, appropriate design of augmentative and alternative communication support can be established, demonstrating positive gains in communication skills such as turn taking, requesting and commenting, as well as increased development of receptive vocabulary, morphology and phonological awareness.

Future research

The aim for further research is to extend the number of items pertaining to other sentence types (such as reversible sentences, 'which' questions, passive sentences, and sentences referring to colours and numerals) in order to evaluate these sentence types in non-speaking children with severe CP.

Acknowledgements

The project (No. 2008043) was financially supported by the Phelps Foundation and the Jack Rabbit Foundation. The authors are grateful for the cooperation of kindergartens and primary schools throughout the Netherlands. The authors express special thanks to all children and their parents who participated in this study. They are grateful for the cooperation of rehabilitation centres, schools for special education and daycare centres throughout the Netherlands.
Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

References

Andersen, G. L., Irgens, L. M., Haagaas, I., Skranes, J. S., Meberg, A. E. and Vik, T., 2008, Cerebral palsy in Norway: prevalence, subtypes and severity. *European Journal of Paediatric Neurology*, **12**, 4–13.

Barbas, H., Garcia-Cabezas, M. A. and Zikopoulos, B., 2013, Frontal–thalamic circuits associated with language. *Brain and Language*, **126**, 49–61.

Binder, J. R., Desai, R. H., Graves, W. W. and Conant, L. L., 2009, Where is the semantic system? A critical review and meta-analysis of 120 functional neuroimaging studies. *Cerebral Cortex*, **19**, 2767–2796.

Bishop, D. V., Brown, B. B. and Robson, J., 1990, The relationship between phoneme discrimination, speech production, and language comprehension in cerebral-palsied individuals. *Journal of Speech and Hearing Research*, **33**, 210–219.

Bruno, J. and Trembath, D., 2006, Use of aided language stimulation to improve syntactic performance during a weeklong intervention program. *Augmentative and Alternative Communication*, **22**, 300–313.

Chater, N. and Christiansen, M. H., 2010, Language acquisition meets language evolution. *Cognitive Science*, **34**, 1131–1157.

Clark, E. V., 2004, How language acquisition builds on cognitive development. *Trends in Cognitive Science*, **8**, 472–478.

Drager, K., Light, J. and McNaughton, D., 2010, Effects of AAC interventions on communication and language for young children with complex communication needs. *Journal of Pediatric Rehabilitation Medicine*, **3**, 303–310.

Dronkers, H. F., Wilkins, D. P., Van Valin, R. D. J., Redfern, B. B. and Jaeger, J. J., 2004, Lesion analysis of the brain areas involved in language comprehension. *Cognition*, **92**, 145–177.

Eliasson, A. C., Krumlinde-Sundholm, L., Rosblad, B., Beckung, E., Arner, M., Ohrvall, A. M. and Rosenbaum, P., 2006, The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. *Developmental Medicine and Child Neurology*, **48**, 549–554.

Falkman, K. W., Sandberg, A. D. and Hjelmquist, E., 2002, Preferred communication modes: prelinguistic and linguistic communication in non-speaking preschool children with cerebral palsy. *International Journal of Language and Communication Disorders*, **37**, 59–68.

Friederici, A. D., 2002, Towards a neural basis of auditory sentence processing. *Trends in Cognitive Science*, **6**, 78–84.

Gainsborough, M., Surman, G., Maestri, G., Colver, A. and Cans, C., 2008, Validity and reliability of the guidelines of the Surveillance of Cerebral Palsy in Europe for the classification of cerebral palsy. *Developmental Medicine and Child Neurology*, **50**, 828–831.

Geytenbeek, J. J. M., Mokkink, L. W., Knol, D. L., Vermeulen, R. J. and Oostrom, K. J., 2014, Reliability and validity of the C-BiLLT: a new instrument to assess comprehension of spoken language in young children with CP and complex communication needs. *Augmentative and Alternative Communication*, **30**, 1–15.

Geytenbeek, J., Harlaar, L., Stam, M., Ket, H., Becher, J. G., Oostrom, K. and Vermeulen, R., J., 2010a, Utility of language comprehension tests for unintelligible or non-speaking children with cerebral palsy: a systematic review. *Developmental Medicine and Child Neurology*, **52**, e267–e277.

Geytenbeek, J. J., Heim, M. M., Vermeulen, R. J. and Oostrom, K. J., 2010b, Assessing comprehension of spoken language in nonspeaking children with cerebral palsy: application of a

newly developed computer-based instrument. *Augmentative and Alternative Communication*, **26**, 97–107.
Harris, C. L., 2003, *Language and Cognition. Encyclopedia of Cognitive Science* (London: Macmillan).
Heim, M. J. M., 2001, *Nauwelijks sprekend veel te zeggen. Een studie naar de effecten van het COCP-programma* [Barely Speaking a Lot to Say. A Study on the Impact of the COCP Programme] (Utrecht: LOT).
Hoff, E., 2009, *Language Development*, 4th edn (Belmont, CA, Wadsworth Cencage Learning).
Hustadt, K. C., Allison, K., Mcfadd, E. and Riehle, K., 2014, Speech and language development in 2-year-old children with cerebral palsy. *Developmental Neurorehabilitation*, **17**, 167–175.
Krageloh-Mann, I., Helber, A., Mader, I., Staudt, M., Wolff, M., Groenendaal, F. and Devries, L., 2002, Bilateral lesions of thalamus and basal ganglia: origin and outcome. *Developmental Medicine and Child Neurology*, **44**, 477–484.
Krageloh-Mann, I., Toft, P., Lunding, J., Andresen, J., Pryds, O. and Lou, H. C., 1999, Brain lesions in preterms: origin, consequences and compensation. *Acta Paediatrica*, **88**, 897–908.
Light, J., 1997, Communication is the essence of human life; reflections on communicative competence. *Augmentative and Alternative Communication*, **13**, 61–70.
Light, J. and Kelford Smith, A., 1993, Home literacy experiences of preschoolers who use AAC systems and of their nondisabled peers. *Augmentative and Alternative Communication*, **9**, 10–25.
Light, J. and Mcnaughton, D., 2011, Supporting the communication, language, and literacy development of children with complex communication needs: state of the science and future research priorities. *Assistive Technology*, **24**, 34–44.
Mercuri, E., Rutherford, M., Barnett, A., Foglia, C., Haataja, L., Counsell, S., Cowan, F. and Dubowitz, L., 2002, MRI lesions and infants with neonatal encephalopathy. Is the Apgar score predictive? *Neuropediatrics*, **33**, 150–156.
Northam, G. B., Liegeois, F., Tournier, J. D., Croft, L., J., Johns, P., N., Chong, W., K., Wyatt, J., S. and Baldeweg, T., 2012, Interhemispheric temporal lobe connectivity predicts language impairment in adolescents born preterm. *Brain*, **135**, 3781–3798.
Palisano, R. J., Hanna, S. E., Rosenbaum, P. L., Russell, D. J., Walter, S. D., Wood, E. P., Raina, P. S. and Galuppi, B. E., 2000, Validation of a model of gross motor function for children with cerebral palsy. *Physical Therapy*, **80**, 974–985.
Parkes, J., Hill, N., Platt, M. J. and Donnelly, C., 2010, Oromotor dysfunction and communication impairments in children with cerebral palsy: a register study. *Developmental Medicine and Child Neurology*, **52**, 1113–1119.
Pennington, L. and Mcconachie, H., 1999, Mother–child interaction revisited: communication with non-speaking physically disabled children. *International Journal of Language and Communication Disorders*, **34**, 391–416.
Perani, D., Saccuman, M. C., Scifo, P., Anwander, A., Spada, D., Baldoli, C., Poloniato, A., Lohmann, G., and Friederici, A.D., 2011, Neural language networks at birth. *Proceedings of the National Academy of Sciences*, **108**, 16056–16061.
Pirila, S., Van Der, M. J., Pentikainen, T., Ruusu-Niemi, P., Korpela, R., Kilpinen, J. and Nieminen, P., 2007, Language and motor speech skills in children with cerebral palsy. *Journal of Communication Disorders*, **40**, 116–128.
Price, C., J., 2012, A review and synthesis of the first 20 years of PET and fMRI studies of heard speech, spoken language and reading. *Neuroimage*, **62**, 816–847.
Rosenbaum, P., Paneth, N., Leviton, A., Goldstein, M., Bax, M., Damiano, D., Dan, B. and Jacobsson, B., 2007, A report: the definition and classification of cerebral palsy April 2006. *Developmental Medicine and Child Neurology*, **109**(**Suppl.**), 8–14.
Ross, B. and Cress, C., J., 2006, Comparison of standardized assessments for cognitive and receptive communication skills in young children with complex communication needs. *Augmentative and Alternative Communication*, **22**, 100–111.
Sevcik, R. A., 2006, Comprehension: an overlooked component in augmented language development, *Disability Rehabilitation*, **28**, 159–167.
Sigurdardottir, S., Eiriksdottir, A., Gunnarsdottir, E., Meintema, M., Arnadottir, U. and Vik, T., 2008, Cognitive profile in young Icelandic children with cerebral palsy. *Developmental Medicine and Child Neurology*, **50**, 357–362.
Sigurdardottir, S. and Vik, T., 2011, Speech, expressive language, and verbal cognition of preschool children with cerebral palsy in Iceland. *Developmental Medicine and Child Neurology*, **53**, 74–80.
Straube, B., Green, A., Weis, S. and Kircher, T., 2012, A supramodal neural network for speech and gesture semantics: an fMRI study. *PloS One*, **7**, e51207.
Turken, A. U. and Dronkers, N. F., 2011, The neural architecture of the language comprehension network: converging evidence from lesion and connectivity analyses. *Frontier Systematic Neuroscience*, **5**, 1–20.
Van Schie, P. E., Siebes, R. C., Dallmeijer A. J., Schuengel C., Smits, D. W., Gorter J. W. and Becher J. G., 2013, Development of social functioning and communication in school-aged (5–9 years) children with cerebral palsy. *Research Developmental Disabilities*, **34**, 4485–4494.
Vasilyeva, M., Waterfall, H. and Huttenlocher, J., 2008, Emergence of syntax: commonalities and differences across children. *Developmental Science*, **11**, 84–97.
Volpe, J. J., 2009, Brain injury in premature infants: a complex amalgam of destructive and developmental disturbances. *Lancet Neurology*, **8**, 110–124.
Voorman, J. M., Dallmeijer, A. J., Van, E. M., Schuengel, C. and Becher, J. G., 2010, Social functioning and communication in children with cerebral palsy: association with disease characteristics and personal and environmental factors. *Developmental Medicine and Child Neurology*, **52**, 441–447.
Vos, R. C., Dallmeijer, A. J., Verhoef, M., Van Schie, P. E., Voorman, J. M., Wiegerink, D. J., Geytenbeek, J. J., Roebroeck, M. E. and Becher, J. G., 2014, Developmental trajectories of receptive and expressive communication in children and young adults with cerebral palsy. *Developmental Medicine and Child Neurology*, **29**, 951–959.
Wassenberg, R., Hurks, P. P., Hendriksen, J. G., Feron, F. J., Meijs, C. J., Vles, J. S. and Jolles, J., 2008, Age-related improvement in complex language comprehension: results of a cross-sectional study with 361 children aged 5 to 15. *Journal of Clinical Experimental Neuropsychology*, **30**, 435–448.
Yin, F. R., Guppy, M. and Johnston, L. M., 2013, Intelligence assessments for children with cerebral palsy: a systematic review. *Developmental Medicine and Child Neurology*, **55**, 911–918.
Zambrani, I. M., Ystrom, E. and Pons, F., 2012, Impact of gender, maternal education, and birth order on the development of language comprehension: a longitudinal study from 18 to 36 months of age. *Developmental Behavioural Pediatrics*, **33**, 146–155.

Appendix A

Table A1. Levels of the GMFCS and MACS

	GMFCS	MACS
Level	Mobility	Handling objects
I	Walks without limitations	Handles objects easily and successfully
II	Walks with limitations	Handles most objects but with somewhat reduced quality and/or spread of achievement
III	Walks using a hand-held mobility device	Handles objects with difficulty; needs help to prepare and/or modify activities
IV	Self-mobility with limitations; may use powered mobility	Handles a limited selection of easily managed objects in adapted situations
V	Transported in a manual wheelchair	Does not handle objects and has severely limited ability to perform even simple actions

Note: GMFCS = gross motor function classification system; MACS = manual ability classification system.

Appendix B: Computer-Based Instrument for Low motor Language Testing (C-BiLLT)

The C-BiLLT consists of a pre-test, a learning module and a computer test:

The pre-test is used to explore whether a child is able to communicate a choice between two concrete objects (or digital photographs of the objects) when provided with the spoken name of the objects. Eight preselected familiar objects (as identified by parents) from the child's home environment (i.e., the child's milk bottle or cup, ball, spoon, coat, trousers, toy car or doll, favourite book, and favourite digital video disc cover) are presented in pairs to the child. While holding a target and foil object in separate hands, the examiner verbally asks the child to select the target object (e.g. 'Where is the ball?') by reaching, pointing, eye-gazing to his/her object of choice. This is repeated in fixed order until all eight pairs of objects have been presented. Next, the child is presented with the same eight objects but now presented as generic photographs of the item (photographic print size A5). Again in fixed order of eight pairs, the child is asked verbally to select the target item. Although the investigator was allowed to encourage the child to provide a response in any way possible, the actual question could only be repeated once. The response of the children was considered correct if the child (based on Heim 2001) fixated his/her eyes on the targeted object for at least 2 s; or reached for the targeted object with his/her arm, foot, elbow or hand; or pointed to the targeted object with his/her hand, arm, foot or head; or turned his/her head to the targeted object with an accompanying vocal sound. If the child correctly identified at least five objects and/or five photographs, he/she was considered able to communicate a selection in response to a spoken response opportunity and advanced to the learning module.

The learning module is incorporated in the assessment package for two reasons. First, to make the child aware of the association between the access methods and the visual representations (response selection) on the 19-inch touch screen. Second, to find empirically the best response mode for the individual child (e.g. eye gazing, touch-screen, input switches or the child's own wheelchair head support) when there are concerns regarding the access method for the child.

The computer test consists of 75 items presented in two parts. Part I consists of three primary sections, with each section containing 10 items referring to nouns, verbs, animals, objects and persons. For each item, the child is shown two digital photographs (a target and a foil) arranged horizontally on the computer screen. To control for effects of chance for each of these three sections, a parallel section exists, presenting the same test items in a different order and with a different foil for each target item (Figure B1). The examiner *always completes* the primary form. Only when a child delivers one or more incorrect responses on the primary form, the examiner assesses the *entire* parallel form.

Part II consists of 45 test items pertaining to spoken sentences with increasing complexity of grammatical structures and is organized into eight sections. The response options per test item are now represented by four digital photographs in a 2 × 2 matrix on the computer screen (Figure B2), providing one target and three foils. For both the first and second parts of the computer test, visual feedback of the child's response is shown by the appearance of a red square around the chosen photograph.

Score calculation

Pre-test (maximum of two points). The child can attain a pre-test score of either one point (for the

Figure B1. Example of an item of the C-BiLLT (test Part I and parallel section): the child is asked 'Where is the shoe ?'

Figure B2. Example of an item of the C-BiLLT (test Part II): the child is asked to identify 'The book is standing on the plate'

identification of at least five real objects *or* five photographs) or two points (for identification of at least five of *both* real objects and photographs). The learning module activities do not contribute to the final score.

Part I (maximum of 30 points). For the computer test Part I (sections 1–3) one point is given for each correctly identified item in both the original and the parallel version with a total of 30 points. For instance if a child answered the question 'Where is the telephone' incorrectly in the primary form and correctly in the parallel form, no point is scored for the item 'telephone'. If the child answered this question correctly both in the primary as well as in the parallel form, one point is scored for the item 'telephone'.

Part II (maximum of 45 points). For the computer test Part II (sections 4–11), one point is scored for each correct response with a total of 45 points.

The maximum achievable C-BiLLT score is 77, i.e. the sum of the pre-test and the scores of the two parts in the computer test.

Figure B1. Example of an item of the C-BiLLT (test Part I and parallel section): the child is asked 'Where is the shoe ?'

Figure B2. Example of an item of the C-BiLLT (test Part II): the child is asked to identify 'The book is standing on the plate'

Figure C1. Example of a noun phrase (NP). The child is asked: '*Waar is de schoen?*' (Where is the shoe?)

Figure C2. Example of a verb phrase (VP). The child is asked '*Wie is aan het slapen?*' (Who is sleeping?)

Appendix C: Examples of visual presentation of the different sentence types

Figure C1. Example of a noun phrase (NP). The child is asked: '*Waar is de schoen?*' (Where is the shoe?)

Figure C2. Example of a verb phrase (VP). The child is asked '*Wie is aan het slapen?*' (Who is sleeping?)

Figure C3. Example of a Who-question? (WhQ). The child is asked '*Wie maakt er muziek?*' (Who is making music?)

Figure C4. Example of a spatial relation embedded in simple sentences (SR). The child is asked to identify the image the test leader is saying: '*de hond ligt in de mand*' (The dog lies *in* the basket)

Figure C5. Example of simple sentences referring to non-observable situations (SOOC) of four persons. The child is asked to identify the image the test leader is saying: '*Joost en Kim gaan buiten spelen, wie blijft er bij mama?*' (Joshua and Kim are going to play outside, who stays with Mammy?)

Figure C3. Example of a Who-question? (WhQ). The child is asked ‘*Wie maakt er muziek?*’ (Who is making music?)

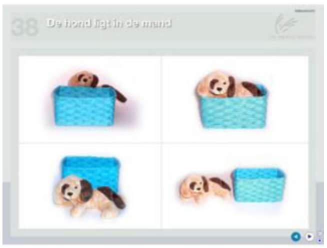


Figure C4. Example of a spatial relation embedded in simple sentences (SR). The child is asked to identify the image the test leader is saying: ‘*de hond ligt in de mand*’ (The dog lies *in* the basket)

Figure C6. Example of a simple sentence with function words (SSF). The child is asked to identify the image the test leader is saying: ‘*Een kleine pot jam staat naast de rode pot jam*’ (A little jar of jam is standing beside the red jar of jam)

Figure C7. Example of compound sentence (CS) referring to the lunch table. The child is asked to identify the image the test leader is saying: ‘*Eerst lag er een appel op tafel maar nu is er een banaan voor in de plaats gekomen*’ (First an apple was lying on the table, but now there is a banana in its place)

Figure C5. Example of simple sentences referring to non-observable situations (SOOC) of four persons. The child is asked to identify the image the test leader is saying: ‘*Joost en Kim gaan buiten spelen, wie blijft er bij mama?*’ (Joshua and Kim are going to play outside, who stays with Mammy?)

Figure C6. Example of a simple sentence with function words (SSF). The child is asked to identify the image the test leader is saying: ‘*Een kleine pot jam staat naast de rode pot jam*’ (A little jar of jam is standing beside the red jar of jam)

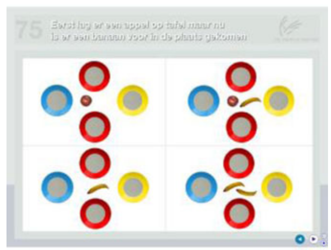


Figure C7. Example of compound sentence (CS) referring to the lunch table. The child is asked to identify the image the test leader is saying: ‘*Eerst lag er een appel op tafel maar nu is er een banaan voor in de plaats gekomen*’ (First an apple was lying on the table, but now there is a banana in its place)