UNIVERSITY OF UTRECHT

MASTER THESIS

Clinical Variables And Language Outcome After Hemispherectomy

A Review Of 20 Cases

Lieselotte Smets
3321576

August 2010

Supervisors:
Dr. Stella de Bode (UMC Utrecht)
Dr. Frank Wijnen (UiL-OTS)

Second reader:
Dr. Sergey Avrutin (UiL-OTS)

ABSTRACT

Research in the field of outcomes of syntactic abilities in patients after cerebral hemispherectomy (i.e. complete disconnection and/or removal of one cerebral hemisphere due to drug-resistant seizures) has not resulted in conclusive findings. A few studies that have been carried out have not reported uniform findings, and it remains unclear what effect on language the removal of left (language-dominant) or right hemispheres is. Important limitations of all existing studies are small numbers of patients (except for one study by Liégeois 2008b) and the use of tests that are not sufficiently grounded in a current linguistic theory. Our first task was to unify and critically assess previous literature. In total, fifteen studies have been reviewed in terms of syntactic tests used and clinical variables such as aetiology reported. Eight studies (total number of patients, n = 14) qualified and their findings are summarized in Figures 1 – 3.

A review of literature suggests that aetiology may be an important factor to take into account in addition to side removed similar to hypotheses that have been put forward by (Curtiss et al. 2001, Liégeois 2008b).

To further investigate this hypothesis we designed a study that partially overcomes limitations of previous reports. We investigated the effect of aetiology x side interactions in a relatively large population of hemispherectomized individuals (n = 20) with well-defined aetiologies limited to two major groups based on the timing of primary insult. We further employed three tests of syntactic ability (ranging from a focus on core syntactic elements to more general grammatical development). Our fourth test addressed lexical comprehension (receptive vocabulary). It was included to examine whether effect of cerebral hemispherectomy on core hard-wired aspects of language are similar in a domain of language that is more influenced by general cognitive abilities (such as IQ).

As a result of the methodological improvements made in this study, we were able to show that aetiology x side do predict outcomes when taken into account together. On all our tests the group with either right or left hemisphere remained perform similarly, but *only* if their primary insult was prenatal. This suggests that a newborn's brain possesses some degree of equipotentiality when it comes to language acquisition. However, even a very short (one year) period of normal development eliminates this equipotentiality. On all tests participants whose lesion occurred after birth performed similarly to adult stroke patients in that the isolated left hemisphere outperformed its right counterpart. This difference in the pattern of language development following hemispherectomy was not limited to syntactic tests only. The same trend was present in the test of receptive vocabulary suggesting that the hemispherectomy effects are not limited to strictly computational aspects of language (such as syntax).

Table Of Contents

1. Introduction

A hemispherectomy is a surgical procedure whereby most cerebral tissue in one hemisphere is resected or disconnected from the rest of the brain. Several types of such surgery exist, and they vary in different ways: not only with respect to the extent of the removal as in anatomic versus functional hemispherectomy (removing the complete hemisphere or only certain lobes), but also regarding which specific layers are removed or left intact (for example, removing only grey matter) (Adams 1983, Carson et al. 1996). Hemispherectomy is considered for patients suffering intractable seizures that are restricted to a single hemisphere, which are caused by a range of unrelated and rare aetiologies, including Rasmussen's encephalitis, Sturge-Weber syndrome and infarcts[1]. Advances in the field of medicine have led to more accurate and earlier detection of such aetiologies, allowing earlier surgery. Hemispherectomies are indeed most often performed on infants (Peacock 1995); many are even carried out before 2 years of age. There is in fact a high success rate for seizure relief in such young children (Duchowny 1993, Mathern et al. 1999, Martínez-González et al. 2005), but also older children and even young adults may benefit from a hemispherectomy too (Telfeian et al. 2002). In contrast, the benefit of hemispherectomy for cognitive functions is more controversial (Pulsifer et al. 2004).

Investigating outcomes of hemispherectomy may have bearing on current scientific research studying a working isolated hemisphere. In these patients, all post-surgical capabilities and any post-surgical development must be housed by the remaining hemisphere. Although the development of patients with unilateral brain lesions is also often studied to identify the potential of a single (non-damaged) hemisphere, there is one empirical problem: in this population, it is unknown whether abilities lost by the damage are compensated for by surrounding cerebral tissue (or other areas in the same hemisphere) or by homologue (or other) areas in the other hemisphere.

[1] Infarcts in the entire thesis refer to prenatal infarcts only (as opposed to postnatal infarcts).

For linguists, more concretely, what is of interest is the post-surgical development of linguistic functions that are typically regarded as strongly lateralised. Syntax and other computational aspects of language are typically dominant in the left hemisphere, whereas other functions are regarded as being more distributed across both hemispheres. Research in this field in the past two decades has not provided uniform or conclusive results. Some studies have found good outcomes in left hemispherectomy patients (i.e. with a remaining right hemisphere) (Mariotti et al. 1998), whereas others have found much poorer results for similar cases of patients (Stark et al. 1995, Ménard et al. 2000). Similarly, both good (Chiricozzi et al. 2005) and poor (Curtiss and de Bode 1998, 1999) results have been found for right hemispherectomy patients. Many larger studies with both left and right hemispherectomy patients also find mixed results (Pulsifer et al. 2004, Liégeois et al. 2008b).

This thesis investigates the current literature of the field, bearing on syntactic outcomes in patients post-hemispherectomy. The number of patients involved in it is small and the tests employed may not always be very good measures of pure syntactic ability. However, the review does suggest that both side remove and aetiology taken into account together may be able to predict outcomes. The current study looks into these two major variables, with a large number of patients (n = 20) and three tests of syntactic ability (including two purely syntactic tests and one that also partly monitors general linguistic development). The results show that only the interaction of the two variables is significant. This points towards equipotentiality of the hemispheres in very young infants that disappears in normal development, when syntactic ability becomes lateralized to the left hemisphere. The right hemisphere then loses its compensatory abilities. In addition to these two major variables, five minor clinical variables are included, of which only seizure control and age at testing were significant. This study only looks into two out of many aetiologies that give rise to severe epilepsy. A short survey of the data available on these other aetiologies suggests that a more detailed classification of them may be needed to accurately predict syntactic outcome for all patients. However, more research is necessary to confirm these speculations.

2. Literature Review

This section reports a range of previous studies on language development in patients post-hemispherectomy. Although this has been researched from a scientifically linguistic perspective since the 1970s by Dennis and colleagues (Dennis & Kohn 1975, Dennis & Whitaker 1976, Dennis 1980), we chose to include only those eight studies that have been published within the past 15 years, and report clinical variables and syntactic abilities[2]. For a full overview of all studies included, see appendix A.

2.1. Unilateral Lesion Literature

Before moving on to the literature discussing outcomes in patients post-hemispherectomy, we review some relevant and recent literature of the unilaterally brain damaged population. This body of literature is larger overall than the hemispherectomy literature and studies often include larger numbers of patients, increasing reliability. One important similarity is the lack of uniformity in the range of results in both fields.

Still, rather interestingly, many studies that describe syntactic outcome in patients with unilateral brain damage do report the same findings: patients with left hemisphere damage eventually perform at the same level as patients with right hemisphere damage (Feldman et al. 1992, Feldman 1994, Bates et al. 1997, 2001, Reilly et al. 1998, Vicari et al. 2000), even if at earlier stages there may be differences observed between the two groups. But a somewhat smaller proportion of the literature reports differences between patients with early left or right hemisphere damage that persist with age and development (Rankin et al. 1981, Aram et al. 1985, 1986, Ballantyne et al. 1994). In these cases it is often the patients with left hemisphere damage who perform more poorly than patients with right hemisphere damage – though not always (Riva & Cazzaniga 1986).

[2] Receptive rather than productive abilities are reviewed here, as much more receptive tests are used with hemispherectomy patients than tests concentrating on production.

However, since the affected hemisphere is not removed in this population, it must be kept in mind that it remains unclear whether any development that should typically be located in this damaged hemisphere is then handled by the other hemisphere, or whether it makes do with the tissue that is left. Therefore, any predictions concerning the hemispherectomy population cannot be straightforwardly extended to the unilaterally lesioned population.

2.2. Hemispherectomy Literature

In the past 15 years of research of linguistic outcome in patients post-hemispherectomy, a variety of aetiologies and various ages of patients have been under investigation. However, the results have not been conclusive, since many different outcomes were found. Patients with a removed left hemisphere have also been studied more often than patients whose right hemisphere was removed. This particular interest is caused by the clear dominance of the left hemisphere for computational aspects of language such as syntax, introducing the question of how well the remaining right hemisphere can support syntactic development, if at all.

In some cases, the remaining right hemisphere appears to be able to develop syntactic ability rather well. Perhaps the most notorious case is presented by Vargha-Khadem et al. (1997). They report of a boy with Sturge-Weber syndrome who was almost mute before his surgery at chronological age (CA) 8;6. At CA 9, his syntactic abilities as formally assessed by the sentence-picture matching task TROG (Bishop 1982) are placed at a test age (TA) 3;6, and at CA 14;11 he receives a TA of 10. Syntactic development is generally viewed as reaching a ceiling level around CA 9 in normal development (Chomsky 1969)[3], but the TROG is normed for all ages. This indicates that the test also incorporates elements that are not strictly syntactic in nature but may include elements from other linguistic domains that continue to develop after CA 9 (see appendix B for more details on all tests reported). The isolated right hemisphere here exhibits the ability to support syntactic

[3] Many proponents of the generative tradition consider most or all syntactic ability as acquired at CA 5 (Brown 1973, Pinker 1994), but Chomsky (1969) shows that some structures may not be acquired until much later.

development to a large extent, albeit still not to normal levels. This patient's scores are shown in Figure 1.

Mariotti et al. (1998) add support to the previous study. They present a left hemispherectomy patient also diagnosed with Sturge-Weber syndrome, who started linguistic development after surgery at CA 3. When extensively examined at age 20, she presents only minor impairments. The patient's verbal IQ is reported at 91, and her performance IQ at 80 points. On a morphosyntactic comprehension subpart of the B.A.D.A. (Miceli et al. 1994) she scores 95% correct[4]. This score is slightly below the range of normal performance (with a cut-off score of 96.7%), but is reported to be comparable to IQ controls. On a different morphosyntactic sentence comprehension test (Pizzamiglio and Parisi 1970) and an active-passive sentence comprehension test (Schwartz et al. 1980) she also scores below the normality range (91.7% correct with cut-off score 96.7%, and 96.2% correct with cut-off score 97.4% respectively) but again these scores are comparable to IQ controls. Still, this patient's scores are very high without exception, and approach all cut off scores very closely. As all scores obtained by this patient are very similar, only the B.A.D.A. score is represented in Figure 2.

Less clear outcomes of syntactic development supported by the isolated right hemisphere are described by Vanlancker-Sidtis (2004). The patient, whose precise aetiology is unknown, was tested at CA 49;6 (44 years after his surgery). Although his level of linguistic ability prior to surgery is not reported, it is noted that his speech was "severely dysarthric with poor intelligibility" (2004:200). He performs well on the Kempler Comprehension task (Kempler 1986), a sentence-picture matching task, scoring 96%. He performs much worse on the Active-Passive Test (Dennis & Kohn 1985), with scores equivalent to age levels 6 and 8. It must be noted, however, that the author herself challenges this particular test on the grounds of not testing syntactic contrast per se (Vanlancker-Sidtis 2004:206, Van Lancker 2001). By including negative sentences, the test also becomes in part semantic. Due to this patient's age, his scores could not be shown in the figures below.

[4] This score is based on the auditory form of the subtest. A visual counterpart was also used, but disregarded here due to possible visual and reading impairments.

The lowest scores on a syntactic test for a left hemispherectomy patient are reported by Trudeau et al. (2003), who present the case of a 17-year-old English/French bilingual girl with Rasmussen's encephalitis. She developed linguistic abilities normally until CA 5, was a balanced bilingual until her surgery at CA 13, and a similar overall impairment is also noted for both languages post-surgery. On the Tests de Langage Dudley-Delage (TLDD, Dudley & Delage 1980), a French adaptation of the Northwestern Syntax Screening Test (NSST, Lee 1969), this patient performs very poorly: at 66.67% correct, her score is equivalent to kindergarten level. This score is represented in Figure 2.

Only one case study was found that examines patient's syntactic abilities after right hemispherectomy. Chiricozzi et al. (2005) examined a girl at CA 19;6 whose seizures started when she was only several months old and were caused by a prenatal infarct, and had surgery at almost 15 years old. Because of the early onset, this patient has not been able to develop linguistic abilities following a normal development, but her presurgical linguistic level is not reported. Interestingly, the B.A.D.A. (Miceli et al. 1994) was used to assess her language development, which Mariotti et al. (1998) used to test a patient with an isolated right hemisphere (described above). The patient with an isolated left hemisphere scored 98.3%, which lies within the normal range. Her score is represented in Figure 2.

Stark & McGregor (1997) report two girls of very similar age after left and right hemispherectomy, both with Rasmussen's encephalitis. They had undergone surgery at comparable ages also (between CA 4 and 6), and had both followed a course of normal linguistic development until approximately CA 2. On the TROG (Bishop 1982), the patient with an isolated right hemisphere does not score very high, in contrast to the patient with an isolated left hemisphere, who performs almost age-appropriately. On the CYCLE-R (Curtiss & Yamada 1988) test also, the patient who had undergone right hemispherectomy does better than the patient who had a left hemispherectomy. Both patients' scores can be found in Figure 1.

A large range of outcomes is found by Liégeois et al. (2008a). They studied six patients of different aetiologies (2 patients suffering Rasmussen's encephalitis, 2 patients with a stroke, one patient with a cyst and one with hemimegalencephaly; 3 patients with left and 3 with right hemispherectomy). Only the two patients suffering Rasmussen's encephalitis had followed a normal course of linguistic development for the first 3 years of life. Using CELF-R (Semel et al. 2000), they evaluate general receptive language scores and find rather uniform result for the patients with an isolated right hemisphere, whose scores are centred around 2-3S.D. below the mean. A much larger spread is found for the patients with an isolated left hemisphere, with two scores over 3S.D. below and one at almost 1S.D. above the mean. It must be noted, however, that CELF-R is a general test of receptive language, and therefore includes subtests that do not measure syntactic ability. All patients' scores are shown in Figure 3.

Liégeois et al. (2008b) studies an even larger body of patients (n = 30), with 9 different aetiologies. They use TROG (Bishop 1982) to evaluate receptive syntactic abilities, but do not report the outcomes of this test separately. Instead, they report scores composed of several linguistic tasks, not all of which are even syntactic in nature (such as the Word Classes subtest of CELF-R [Semel et al. 2000], which is a semantic association task). They also do not report raw scores or scores by patient or by aetiology, but rather provide significance results for the left versus right patient groups. Raw scores on CELF-R for one patient were obtained (F. Liégeois, pers. comm.) and are represented in Figure 3. Their general findings are that patients with an isolated left hemisphere perform better than patients with an isolated right hemisphere, "but only if onset of pathology was postnatal" (2008b:3104). This conclusion already suggests that the factor of aetiology may play an important role together with side removed in predicting linguistic outcomes post-hemispherectomy.

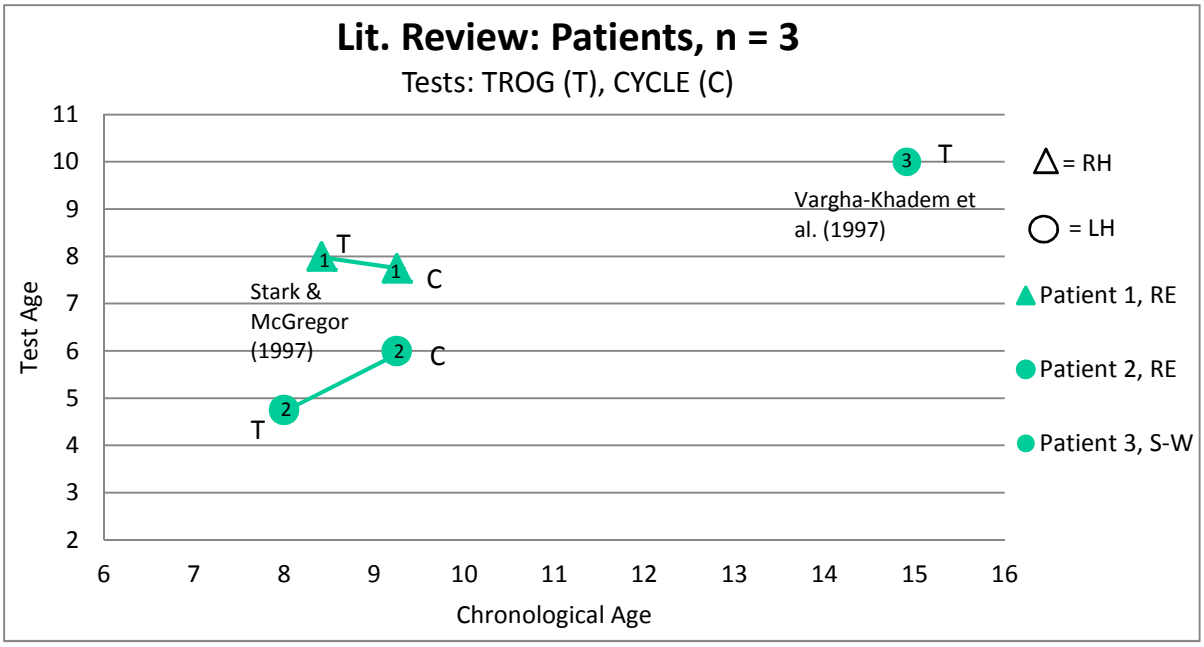


Figure 1. Results for 3 patients on TROG (T) and CYCLE (C), scored in test age by chronological age.
RE = Rasmussen's encephalitis
S-W = Sturge-Weber syndrome

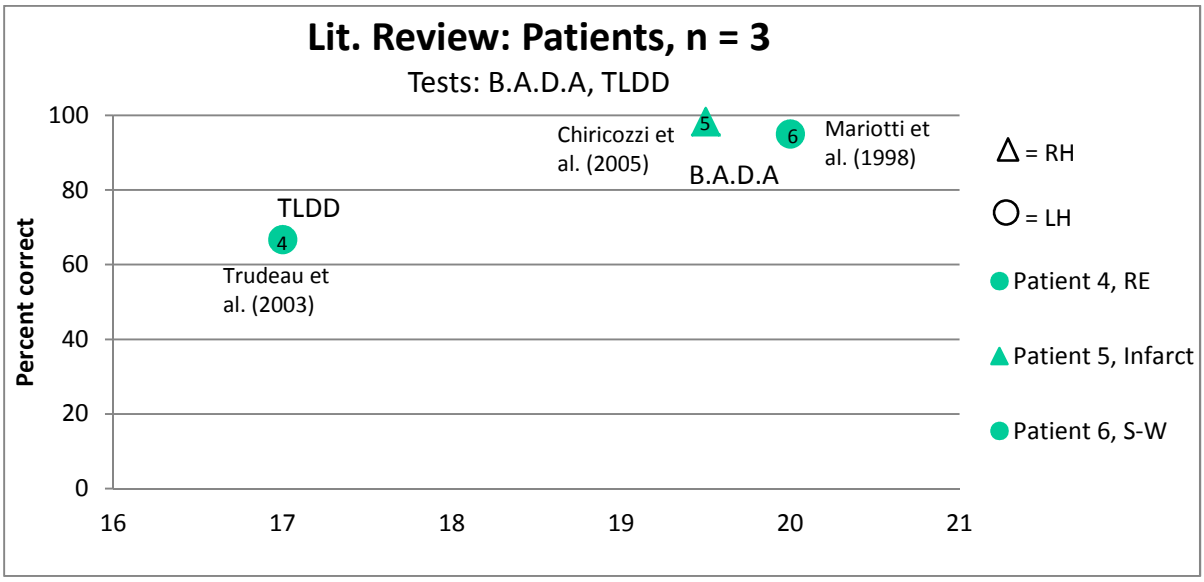


Figure 2. Results for 3 patients on the TLDD and B.A.D.A., scored in percentages correct by chronological age.
RE = Rasmussen's encephalitis
S-W = Sturge-Weber syndrome

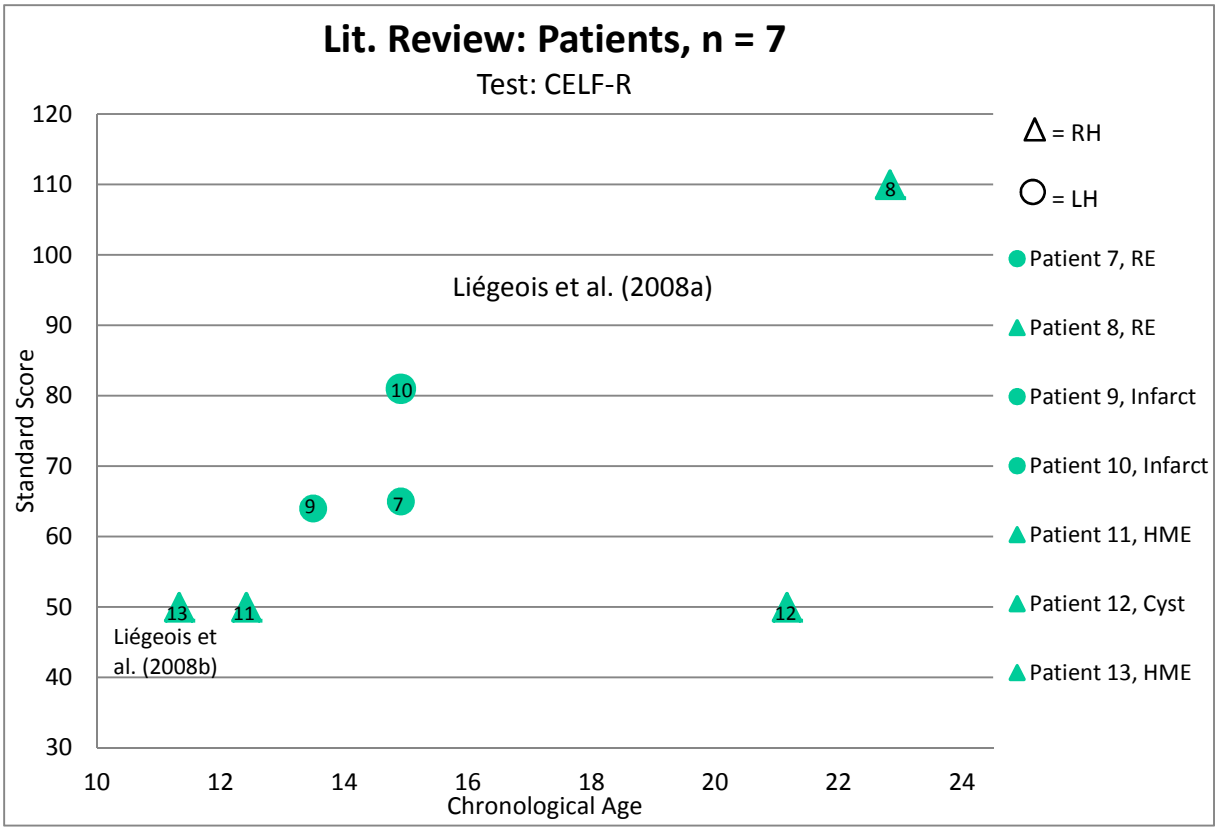


Figure 3. Results for 6 patients on the CELF-R, in standard scores by chronological age.
1S.D. = ±15 points
RE = Rasmussen's encephalitis
HME = hemimegalencephaly

The literature review shows that not only side removed, but also aetiology needs to be taken into account to predict syntactic ability. In particular, the data consistently show that Rasmussen's encephalitis is an aetiology that has very different outcomes depending on which hemisphere is removed. This is in accordance with Curtiss et al. (2001), who found that patients of acquired aetiology (Rasmussen's encephalitis) perform differently from developmental aetiologies (infarcts, Sturge-Weber syndrome, hemimegalencephaly and cysts) in addition to the right versus left distinction. In the data from the literature review, both patients 1 and 8 are right hemispherectomy patients with Rasmussen's encephalitis, and both outperform all other patients – not only left hemispherectomy patients with Rasmussen's encephalitis (patients 2, 4 and 7). Patients with developmental aetiologies can reach high levels regardless of side removed (patients 5 and 6 in

particular, but also patients 3 and 10). Only the two patients with hemimegalencephaly and one with a cyst do not perform well at all.

3. Theoretical Framework

The debate on the innate predisposition of the left hemisphere for language dates back decades, and is one that has still not been resolved. On the one hand, the more traditional view holds that linguistic (in particular syntactic) functions are innately present in the left hemisphere, so that this hemisphere is already predisposed before any acquisition occurs (Molfese & Segalowitz 1988). On the other hand, Lenneberg (1967) has proposed that both hemispheres are equipotent at birth. The brain undergoes no lateralization for linguistic functions until 20 months of age, when it starts developing; though many current proponents accept that general language development starts much earlier than 20 months (Locke 1993). Both views can provide an explanation for why the right hemisphere can also support language acquisition, as is the case in a small proportion of neurologically unimpaired people (Rasmussen & Milner 1977). Under the innate lateralization view, this is possible because of the brain's plasticity and acquisition will be somewhat less efficient than in the left hemisphere (Witelson 1987), whereas in the equipotentiality view this is a natural consequence of the lack of early left lateralization.

Regardless of which view is advocated, the difference between developmental (early onset) and acquired (late onset) aetiologies is crucial to understanding the outcome of patients' syntactic abilities. If the onset is early in the right hemisphere, then both views of lateralisation predict that acquisition should be able to proceed normally in the left hemisphere. If the left hemisphere is affected, then only the equipotentiality view guarantees normal development to take place in the right hemisphere, whereas the lateralisation view claims that acquisition will be effortful and perhaps slower, though (near-)normal levels can still be reached. If the onset of aetiology is late, both views

predict that an affected right hemisphere will not have an impact on syntactic ability, though an affected left hemisphere will severely impair it.

3.1. Hemispherectomy Literature

The data gathered from the literature review for the most part reflect this distinction of aetiology onset. In Figures 1 and 2, all patients with a developmental (early) aetiology acquire syntactic ability to at least near-normal or only mildly impaired levels, as do all patients with an acquired (late) aetiology affecting the right hemisphere. Patients with an acquired aetiology affecting the left hemisphere show much greater impairments. In Figure 3, the patients with an acquired aetiology show the same pattern, but those with a developmental aetiology in fact do not perform as predicted. In particular, patients with an isolated left hemisphere receive the lowest scores – a fact that goes against both views of lateralisation.

Looking at the data more closely, we see that all patients after left hemispherectomy (i.e. with a remaining right hemisphere) perform as predicted. The patients with the acquired aetiology Rasmussen's encephalitis perform poorly: patient 2 receives a score far below her chronological age and is noted to perform like other language impaired children (Stark & McGregor 1997), the score obtained by patient 4 is noted to be equivalent to kindergarten level (Trudeau et al. 2003) and patient 8 scores at over 2S.D. below the norm despite a very long post-surgical recovery period (Liégeois at al. 2008a). As their left hemisphere had already started to acquire syntactic ability normally and therefore become lateralised for it (if not already present at birth), the onset of aetiology and consequent removal have erased most knowledge. The right hemisphere is left to make up for this, but only has reduced plasticity at this age to do so, and therefore does not succeed very well.

Of the patients after left hemispherectomy with developmental aetiologies, there are two patients with Sturge-Weber syndrome. Although they remain impaired to some extent, patients 3 and 6 have made large improvements and the latter is noted to perform similarly to other patients

with equal IQ levels (Mariotti et al. 1998). Out of the two patients who had suffered an infarct, one seems to perform much more poorly than the other, with a full S.D. separating the two. However, patient 10 has only been able to recover from their surgery for a relatively short period of time, which may indicate that post-surgical development is still on-going (Liégeois et al. 2008a). This patient may still reach the same level as patient 9, who is more mildly impaired. At the time of onset, these patients' had either not undergone any lateralisation or possessed a sufficiently plastic right hemisphere that could more easily pick up syntactic ability than that of patients with Rasmussen's encephalitis.

On the other hand, not all patients who have undergone right hemispherectomy appear to perform as predicted above. Those who do perform as predicted are those patients with the acquired aetiology Rasmussen's encephalitis. Both patients 1 and 8 receive scores that are age-appropriate. Indeed, the former is reported to develop similarly to neurologically normal children (Stark & McGregor 1997), and the latter is the only in the sample to score above the norm (though within 1S.D., Liégeois et al. 2008a). These patients' remaining left hemispheres are able to develop normally being or becoming lateralised for computational functions, without being (very) affected by the aetiology in the right hemisphere. The knowledge that has developed in the left hemisphere remains there, and does not need to be compensated for.

Out of the four patients with a developmental aetiology affecting the right hemisphere, there is only one patient has suffered a prenatal infarct. As the typically non-computational hemisphere is involved, syntactic development should not be affected very much, and this is reflected in the high scores obtained by patient 5. The same should hold for patients 11, 12 and 13, of whom two patients have hemimegalencephaly and one has a cyst. However, their performance is in fact very poor as they all receive the lowest score possible on the task. They were tested on a general measure of linguistic ability as mentioned above, but these patients scored poorly on all subtests (F. Liégeois, pers. comm.). This indicates that their overall scores were not pulled down by subtests focusing on

non-computational linguistic functions, and syntactic ability is thus clearly impaired. As the typically non-computational hemisphere is involved (at an early age where plasticity in the brain is maximal), this goes against what is predicted above.

4. Current Study And Hypotheses

The typical lateralisation of syntax to the left hemisphere implies the importance of which side is removed in a hemispherectomy. However, the timing of aetiology onset (developmental versus acquired) is also revealed to be of significance by the literature review. Both factors are then considered to be of importance to accurately predict syntactic ability in patients post-hemispherectomy. The following specific hypotheses arise:

- Infarcts and Sturge-Weber syndrome, both early aetiologies, do not show a difference in outcomes for patients of either side removed.
- Rasmussen's encephalitis, a late aetiology, shows a difference in performance between right and left patients. More specifically, patients after right hemispherectomy are predicted to outperform patients after left hemispherectomy.

We also include other clinical variables (seizure control, medication, ages at seizure onset[5], surgery, and testing) to examine their influence on patients' outcomes in addition to the two main variables of side removed and aetiology. Our study mainly focuses on syntactic ability, but also includes a test on lexical comprehension to measure to what extent these claims also hold for a linguistic domain that is more influenced by general cognitive abilities.

[5] Note that age at seizure onset does not indicate the age of onset of insult. It is often the age at which symptoms are first noticed, but the primary insult may have happened long before this point in time.

5. Methods

The data were collected at the University of South Carolina, USA between 2005 and 2007, as a part of Stella de Bode's NIH-funded grant R 21.

5.1. Participants

5.1.1. Hemispherectomy Patients

All patients who entered rehabilitation at the University of South Carolina between 2005 and 2007 participated in this study, and were monolingual speakers of English. Of the total of twenty patients, eight had undergone a right hemispherectomy (i.e. with a remaining left hemisphere), of which three were diagnosed with an infarct, four with Rasmussen's encephalitis and one with Sturge-Weber syndrome. Twelve patients had undergone a left hemispherectomy (i.e. with a remaining right hemisphere); of which eight have been diagnosed with an infarct, three with Rasmussen's encephalitis and one with Sturge-Weber syndrome. There are only two patients with Sturge-Weber syndrome in total, which we believe to be too few to be representative of the entire population of patients with this aetiology. Therefore, their scores are reported in the figures but not included in the analysis. Patients' age at the time of testing ranged from 5;11 to 25;2 with a mean of 14;6 (S.D. = 5;6). Ages at seizure onset ranged from 0;1 to 12;0 with a mean of 4;4 (S.D. = 3;6). Patients' age at surgery ranged from 1;0 to 15;4 with a mean of 7;5 (S.D. = 4;6). Surgery had brought complete relief of seizures for fifteen patients, of which ten were also no longer taking any medication. Seizures were persisting to some degree for five patients despite still taking medication. An overview of patient information is given in Table 1.

Patient no.	Side removed	Aetiology	Age at testing	Age at seizure onset	Age at surgery	Seizure free	Medication free
H002	R	PI	12;2	7;2	7;5	Y	N
H003	L	PI	13;4	5	8;4	Y	Y
H004	L	PI	14;4	4	10;0	Y	Y
H102	L	PI	21;6	0;6	10;6	Y	Y
H103	R	RE	21;8	10	11;10	Y	N
H104	L	PI	14;0	4;2	6;3	Y	Y
H106	R	PI	17;11	2;5	15;4	Y	N
H107	L	PI	5;11	0;1	1;2	Y	Y
H108	R	RE	5;11	4	4;6	Y	N
H110	L	RE	14;2	2	4;4	N	N
H111	R	PI	20;9	2;6	1;11	N	N
H112	L	PI	9;8	3	4;2	N	N
H113	L	RE	17;4	11	12;1	Y	Y
H114	L	S-W	10;4	0;3	1;0	Y	Y
H115	R	RE	25;2	2;6	3;10	Y	Y
H116	L	PI	20;0	7	11;5	Y	Y
H117	R	S-W	10;3	3	7;4	Y	Y
H118	L	RE	8;9	1	1;11	N	N
H119	L	PI	10;11	5	9;11	Y	N
H120	R	RE	16;7	12	15;4	N	N

Table 1. Overview of patient information. Ages are years;months or only years if number of months is unknown.
PI = prenatal infarct, RE = Rasmussen's encephalitis, S-W = Sturge-Weber syndrome
Y = yes, N = no

5.1.2. Control Subjects

Fourteen control participants also participated in this study. These were all normally developing children between ages 7;2 and 10;4 (mean 8;8, S.D. 0;11). They were all children from the Jewish community in South Carolina to ensure that they natively spoke Standard American English rather than the local dialect commonly spoken by other communities in this area. As the tests target Standard English, the most reliable results are obtained by speakers of this variety of English.

5.2. Materials

Four different linguistic tests were used: three of syntactic nature (Curtiss and Yamada Clinical Language Evaluation [CYCLE], Curtiss & Yamada 1988; Sentence Judgement, unpublished at UCLA; Binding, Control and Presupposition [BCP], unpublished at UCLA), and one test of lexical

comprehension (Peabody Picture Vocabulary Test, Third Edition [PPVT-III], Dunn & Dunn 1997). It must be noted that not all participants were tested on all tasks, though the majority (16 patients) was.

5.2.1. CYCLE

The receptive portion of the CYCLE (Curtiss & Yamada 1988) was used to test patients' syntactic abilities. It is a battery of short tests distributed over 9 age levels, each of which contains tasks that focus on areas of linguistic knowledge that are usually acquired at that age in normally developing children. Age 9 is the ceiling level, and the child is considered to have a mature grammar in place if it passes this level. A level may be passed if the subject passes at least 75% of the subtests, or half-passed if between 50-74% of the subtests are passed. The tasks are in fact not all syntactic in nature, but may also focus on lexical comprehension instead. Still most of the lexical comprehension tasks are included in the earlier age levels, whereas most of the syntactic tasks are included in the later age levels. Since most of the patients are older than 9 years of age (and controls are around this age), we have focused on the later age levels only (ages 6 – 9), making this test appropriate for detecting syntactic ability. Most subtests are sentence-picture matching tasks, though some required the subject to perform a certain action in response to cues given by the examiner. An example of an item from a lexical comprehension task of an early age level is given in (1); an example of an item from a syntactic task of a later age level is given in (2).

(1) Age level 2, Subtest 2.5: *Lexicon: Reference Words II*

Point to the shoe. (2 picture options)

(2) Age level 9, Subtest 9.4: *S-O Relative Clauses*

The girl who the boy is pushing is happy. (4 picture options)

5.2.2. Sentence Judgement

This test (unpublished at UCLA) requires the subject to judge whether a sentence spoken by the examiner is grammatically correct or not. It is assumed that if a subject correctly judges these sentences, they have the syntactic ability required to do so. There are 50 sentences to judge in total, of which half are correct and the other half are not. Twelve syntactic constructions are included, so that each is tested four times; twice grammatically and twice with corresponding ungrammatical sentences. The different constructions are: 2 types of verb subcategorisation, present continuous inflection (BE + -ING), X-bar constructions, subject-verb agreement, subject and object case inflection, tense inflection, *do*-support, wh-questions, negatives, 2 types of subject-auxiliary inversion. Although the task was designed to test syntactic ability, some of the categories are no longer regarded as (morpho-) syntactic in current syntactic theory. Negative constructions and tense inflection in particular are currently regarded as being more semantic than syntactic in nature (see Hornstein et al. 2005). However, only few items are concerned outdated, so the overall validity of the test remains. An example of a grammatically correct sentence and its ungrammatical counterpart are given in (3) and (4).

(3) Does Bill arrive in the morning? (GOOD)

(4) *Arrives Bill in the morning? (BAD)

Syntactic construction: *do*-support

5.2.3. BCP

The Binding, Control and Presupposition test (unpublished at UCLA) is designed to test core syntactic constructions and is composed of three subparts, as indicated by the full title. The Binding subpart tests only principle C from the classic Binding theory (Chomsky 1981) in 4 items; the Control subpart contains 6 items with obligatory control constructions in which principle B is tested; and the Presupposition subpart contains 6 items testing pragmatic knowledge. The items are all presented in

a similar format, where the test sentence is introduced and a question is asked about it to elicit an answer from the subject. All items from the different subparts are presented together in a mixed order, so that the different sentence types are masked. The performance on the first two subparts is measured, and the third merely provides non-syntactic filler items. An example of each subpart is given in (5), (6) and (7), where test sentences are marked in bold.

(5) Binding: *Could John be the one that made lunch? Listen.* ***He made lunch when John got hungry****. Could John be the one that made lunch?* (YES)

(6) Control: *Could Steve be the one that got drawn? Listen.* ***Steve promised Larry to draw him, and he did.*** *Could Steve be the one that got drawn?* (NO)

(7) Presupposition: ***Jane remembered to open the door****. Was Jane supposed to?* (YES)

5.2.4. PPVT

The only test of lexical comprehension that was included is the PPVT-III (Dunn & Dunn 1997). It is composed of seventeen sets of lexical items that need to be matched to the correct pictures. The sets are of increasing difficulty, ranging from a child's typical first words to words that are highly infrequent even among adults. The raw scores obtained may be converted to standard scores in order to compare relative vocabulary size in subjects of different ages, or to equivalent mental ages (among other scoring types). The PPVT-III is normed for all ages (2 – 90+), and is in fact the only test included here which has been normed at all. Examples of one word of each set are given in (8).

(8) Increasing difficulty: *bus, drum, empty, astronaut, delivering, oval, horrified, flamingo, bouquet, inflated, hazardous, pedestrian, syringe, poultry, quintet, coniferous, terpsichorean*

6. Results

6.1. Control Subjects

Control subject generally performed well on the tasks. Eleven out of fourteen subjects (78.6%) passed age-appropriate levels for CYCLE, with some even passing one or two levels higher than their chronological age (Figure 4). Only one subject just reached level 7, performing somewhat below their chronological age.

Concerning the Sentence Judgement[6] task (Figure 5), only one patient judged all sentences correctly, but a total of eleven out of fourteen subjects (78.6%) judged at least 45 sentences (90%) correctly. The three subjects who obtained lower scores had also received the lowest scores on the CYCLE test.

On the only lexical comprehension task, control subjects performed better than average (Figure 6). With the exception of only one subject, all perform at or (far) above the norm; seven subjects (50%) receive scores at over 1S.D. above the norm. The high scores may be due to their education and background. Unlike syntactic ability which relies on an internal computational system, lexical comprehension ability may be influenced by such social factors.

Control subjects in fact did not perform very well on the Binding subtest of the BCP (Figure 7). Only four subjects (28.6%) answered all four items correctly, and five subjects (35.7%) only answered one or two items correctly[6].

[6]The Sentence Judgement and BCP tasks require subjects to give 'yes' or 'no' answers. This means that performances may be influenced by a yes response bias. By calculating d' scores for all patients it was examined if such a trend is indeed present in the data. The higher the d' value, the less likely it is that a bias is present. We take a d' smaller than 1.5 to indicate the presence of a bias. Controls had an average d' of 3.282 for Sentence Judgement, which clearly indicates that there is no bias. For the Binding subtest the average d' is 1.994, also pointing towards the absence of a bias.

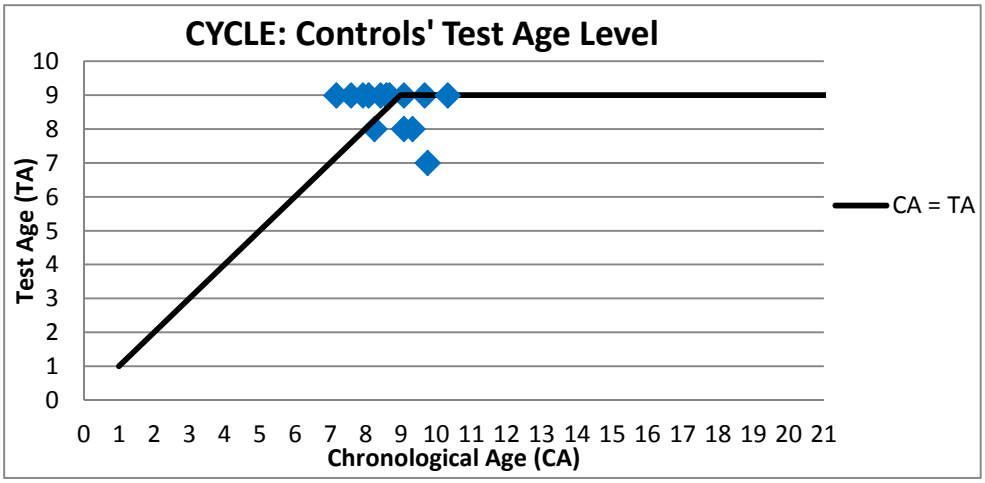


Figure 4. Performance of control subjects on the CYCLE test.

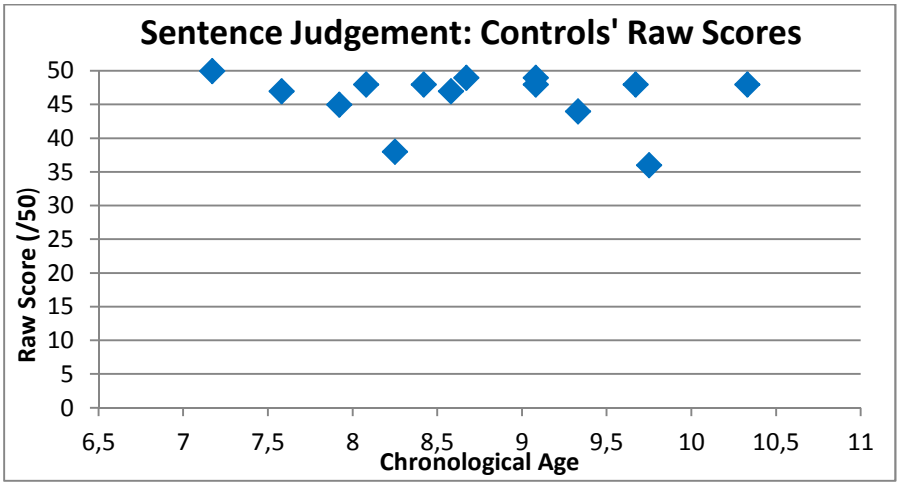


Figure 5. Performance of control subjects on the Sentence Judgement test.

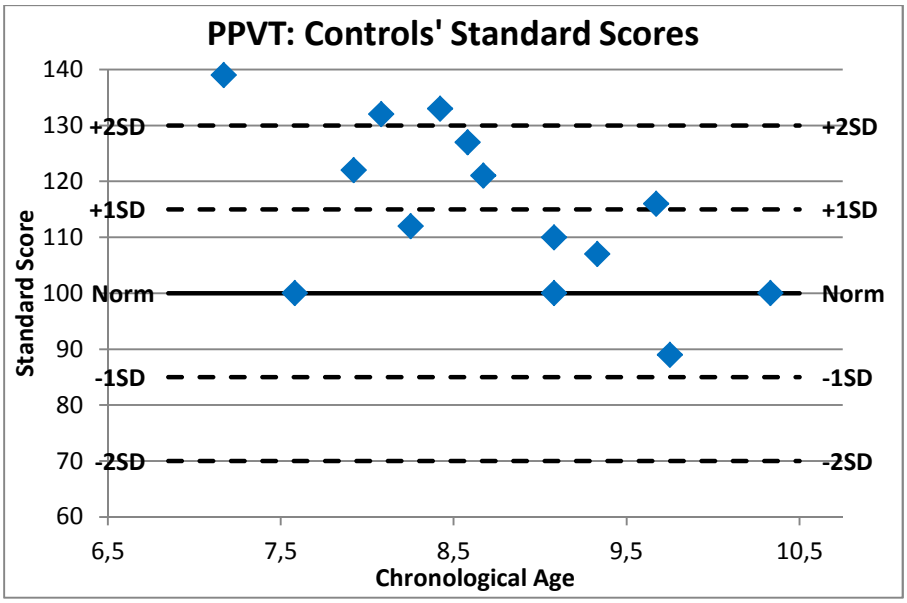


Figure 6. Performance of control subjects on the PPVT.

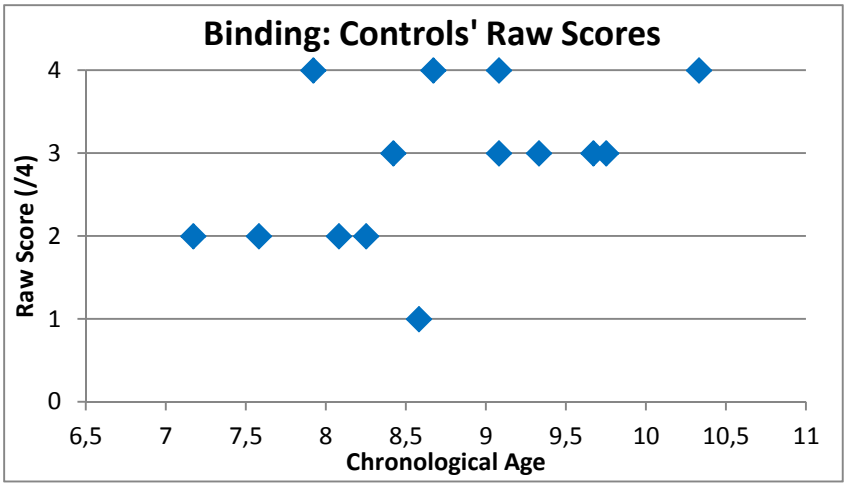


Figure 7. Performance of control subjects on the Binding subpart of the BCP test.

6.2. Hemispherectomy Patients

6.2.1. Major Variables: Side Removed And Aetiology

Nineteen hemispherectomy patients (seventeen without the Sturge-Weber patients) were tested on CYCLE. Only patient H002 did not participate for this test. Sixteen patients were tested on the Sentence Judgement task and the BCP (fourteen without Sturge-Weber patients), as patient H002 again did not participate, and the three youngest patients (H107, H108, H118) did not pass the pre-tests. All twenty patients (eighteen without Sturge-Weber patients) were tested on the PPVT. For the details on all patients, see Table 1 on p. 14.

Similar results were found for the CYCLE, Sentence Judgement[7] and PPVT tests (Table 2). First, we explored the factors of side removed and aetiology separately, and neither of the two variables reached significance. Next, we investigated the interaction between these two variables, which reached significance at the .10 level[8] for CYCLE and Sentence Judgement, but was not significant at the .10 level for PPVT. The observed trend in all three cases indicates no difference between right and left patients with an infarct on the one hand, whereas there is a difference between right and

[7] With an average d' of 2.703 and only two d' scores below 1.5, it is concluded that overall no yes response bias is present for hemispherectomy patients on the Sentence Judgement task.

[8] The significance level may be considered for raising up to .10 because of the small number of patients in each side removed/aetiology group.

left patients with Rasmussen's encephalitis on the other hand. Namely, right patients consistently outperform left patients (Figures 8, 9, and 10).

Test Variable	CYCLE	Sentence Judgement	PPVT	Binding subtest of BCP
Side Removed	.122	.442	.227	.696
Aetiology	.276	.917	.568	.637
Side Removed*Aetiology	***.083***	***.084***	.125	.344

Table 2. Results of the statistical analysis (two-way ANOVA) for the main variables on the four tests. Scores significant at the .10 level are in italics.

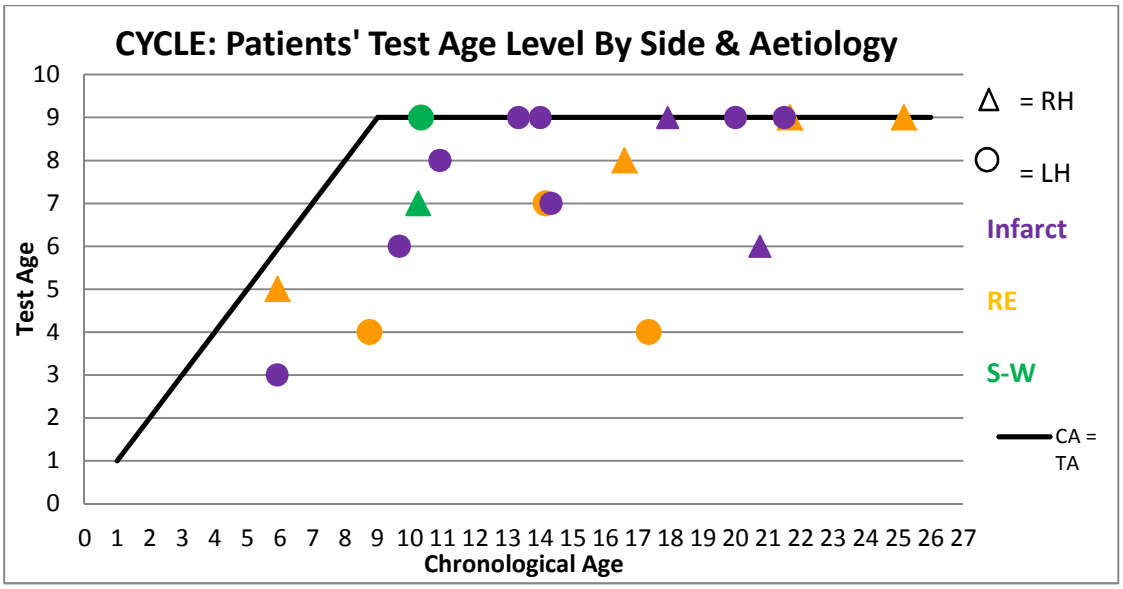


Figure 8. Performance of patients on the CYCLE test, indicating for each patient which side has been removed as well as their aetiology. Note that the scores are represented as Test Ages, whereas the statistical analysis is performed on developmental quotient scores (= test age divided by chronological age).
RE = Rasmussen's encephalitis
S-W = Sturge-Weber syndrome
LH = left hemisphere removed
RH = right hemisphere removed

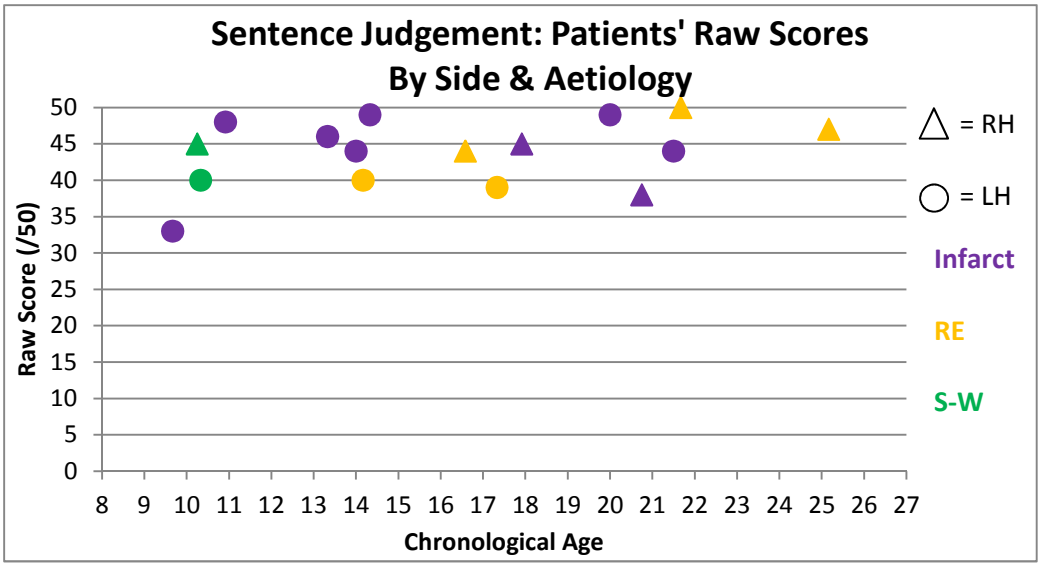


Figure 9. **Performance of patients on the Sentence Judgement test, indicating for each patient which side has been removed as well as their aetiology.**
RE = Rasmussen's encephalitis
S-W = Sturge-Weber syndrome
LH = left hemisphere removed
RH = right hemisphere removed

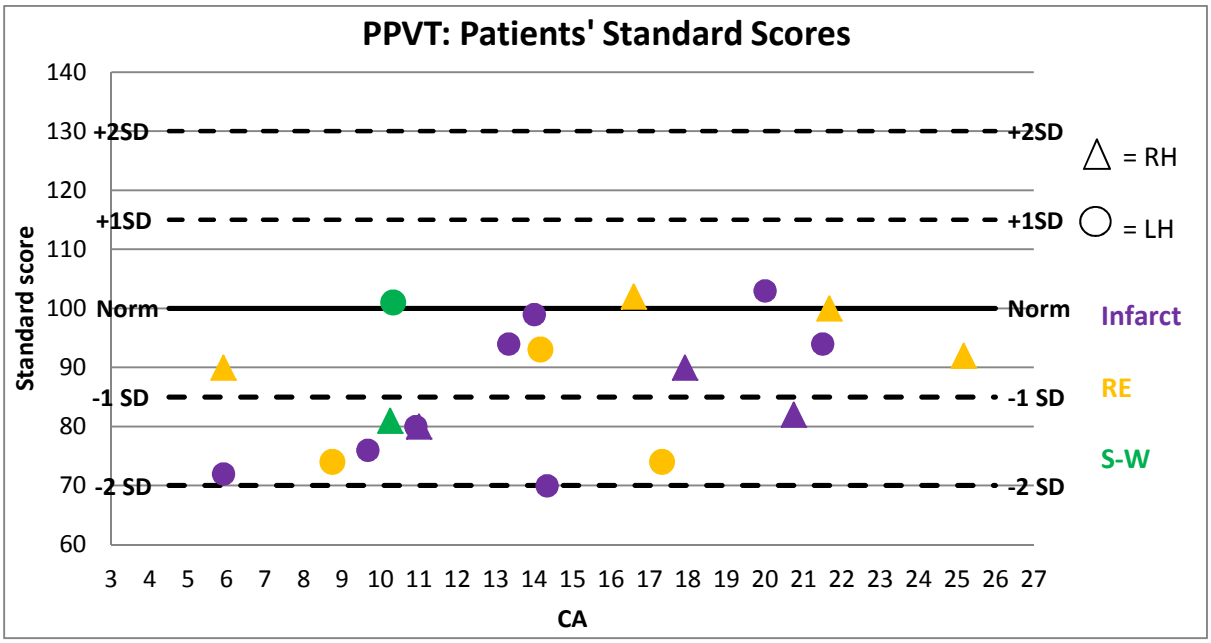


Figure 10. **Performance of patients on the PPVT, indicating for each patient which side has been removed as well as their aetiology.**
RE = Rasmussen's encephalitis
S-W = Sturge-Weber syndrome
LH = left hemisphere removed
RH = right hemisphere removed

Concerning the Binding subtest of the BCP, a yes response bias seems to be present among the patients as indicated by an average d' of 1.241. Only one patient answered all items correctly, and three answered all items with 'yes'. Five patients answered 'yes' three out of four times, and despite the low number of items we were led to assume that this was due to a bias because of their performance on both Binding and Control sentences. Here, patients also showed a yes response bias (average d' = 1.202, four patients answered [almost] all ten items with 'yes').

One explanation for the presence of a yes response bias is the felicity of the test items. Although they are all syntactically correct, they may not be logical pieces of discourse. In particular the items in the Binding subpart may be confusing to participants, as pronouns refer to discourse referents that have not yet been introduced and are therefore not salient from the context. An example is the test sentence reported in the test description, repeated here in (9). The test sentence refers to a discourse entity by using the pronoun *he*, whereas its referent has not been introduced in the discourse, as is normally the case. The subject may then link this entity to the only one that has already been introduced, namely *John*. The inconsistency that arises from a mismatch between syntax and discourse may then give rise to the yes response bias.

(9) Binding: *Could John be the one that made lunch? Listen.* ***He made lunch when John got hungry****. Could John be the one that made lunch?* (YES)

The presence of a bias may explain the pattern of results found when looking into the main variables of side removed and aetiology. Neither of the two separately can explain the results, and the interaction between the variables is also not significant (Table 2). The trend found for the other tests is not present for the Binding subtest (Figure 11).

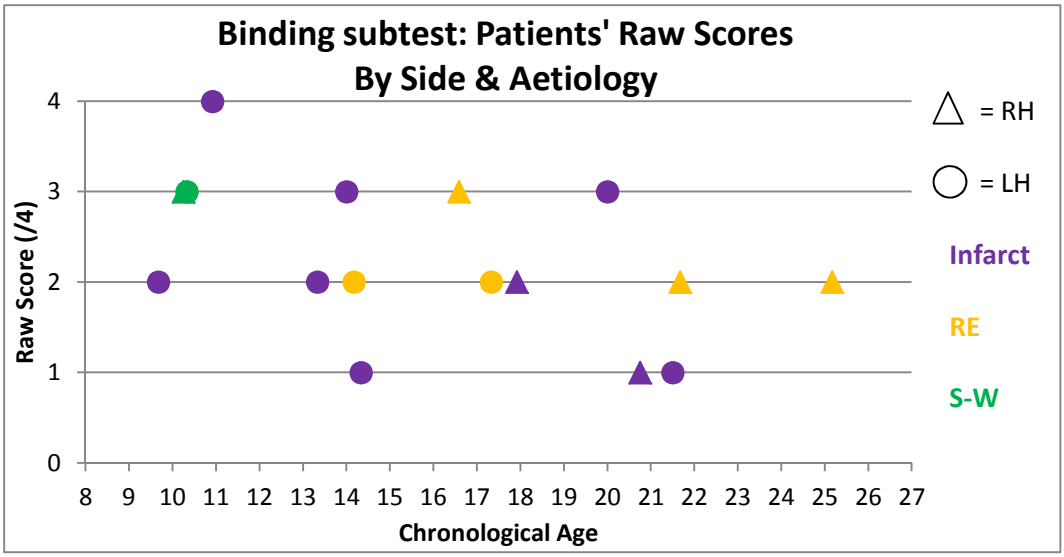


Figure 11. Performance of patients on the Binding subpart of the BCP test, indicating for each patient which side has been removed as well as their aetiology.
RE = Rasmussen's encephalitis
S-W = Sturge-Weber syndrome
LH = left hemisphere removed
RH = right hemisphere removed

Post-hoc comparisons were made for all tests by comparing each side removed/aetiology group. These comparisons revealed the same trend in the test results, with the exception again of the Binding subtest. For CYCLE, Sentence Judgement and PPVT, the difference between right and left patients with an infarct is not significant. On the other hand, the difference between right and left patients with Rasmussen's encephalitis is in all three cases significant. Considering the presence of a bias in the Binding subtest, it is no surprise that the side comparisons are not significant within both aetiologies. An overview of the statistics is given in Table 3, and the averages of all groups are shown per test in Figures 12 – 15.

Test Comparison	CYCLE	Sentence Judgement	PPVT	Binding subtest of BCP
Infarct LH vs RH	.446	.245	.403	.194
Rasmussen's encephalitis LH vs RH	***.007***	***.023***	***.020***	.248

Table 3. **Results of post-hoc comparisons (one-tailed t-test) for the four tests. Significant scores are in bold italics.**

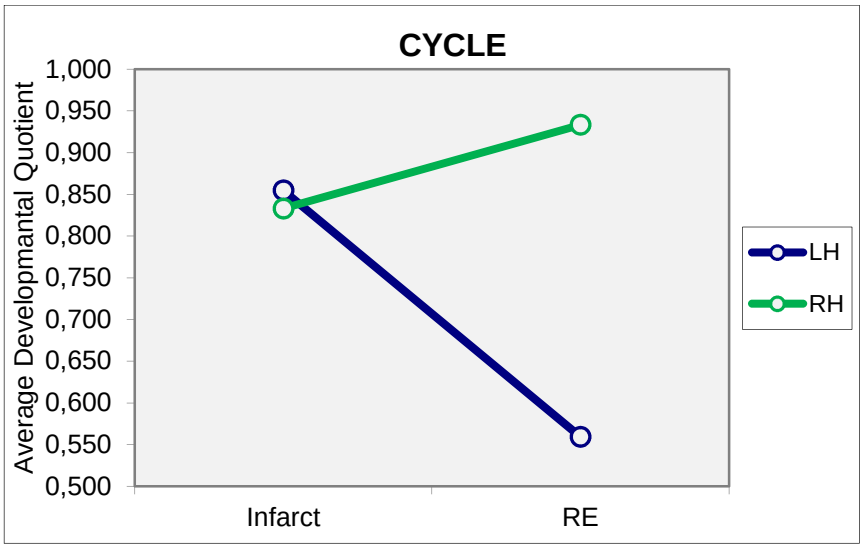


Figure 12. **Averages of developmental quotient per side removed/aetiology group. RE = Rasmussen's encephalitis**

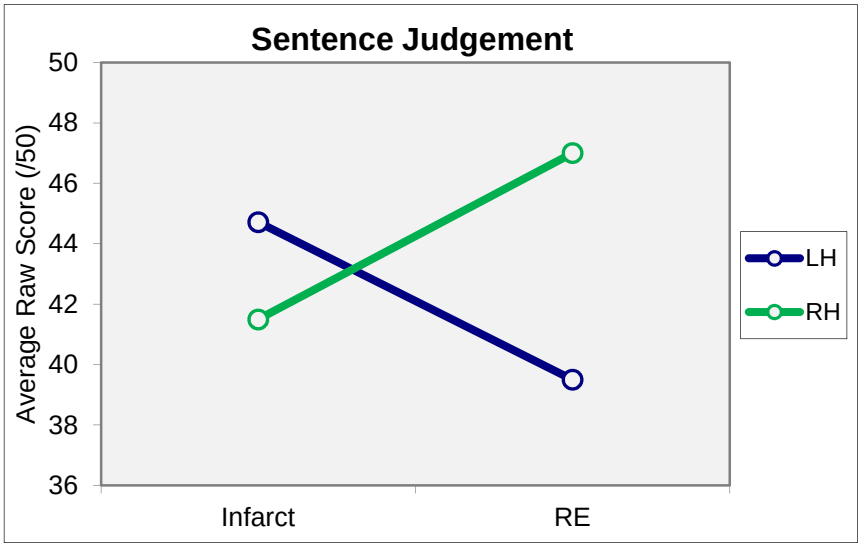


Figure 13. **Averages of raw scores per side removed/aetiology group. RE = Rasmussen's encephalitis**

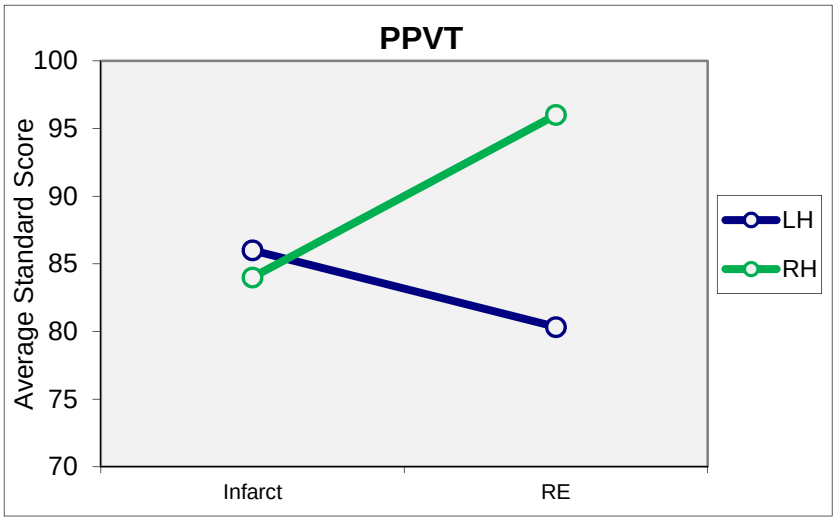


Figure 14. Averages of standard scores per side removed/aetiology group.
RE = Rasmussen's encephalitis

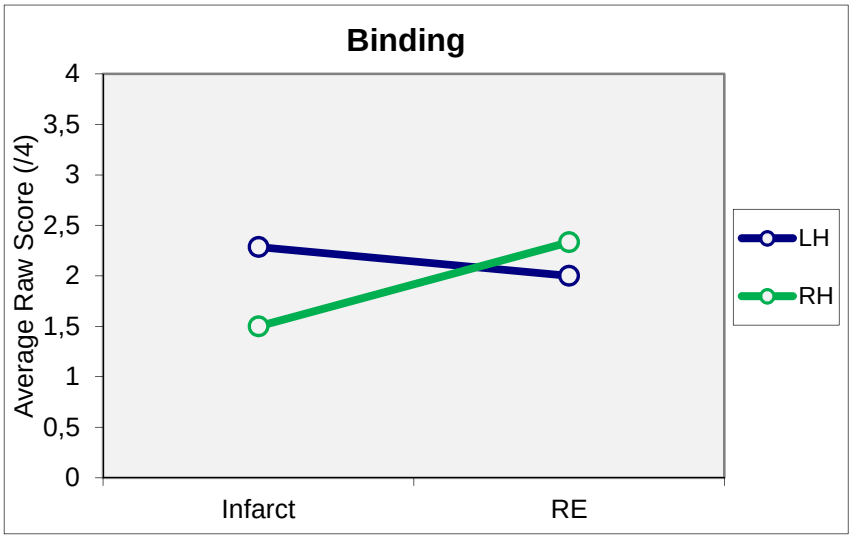


Figure 15. Averages of raw scores per side removed/aetiology group.
RE = Rasmussen's encephalitis

6.2.2. Other Clinical Variables

We also examined the relevance of five additional variables: seizure control, medication, age at seizure onset, age at surgery and age at testing. The variables of medication, age at seizure onset and age at surgery were not significant for any test. Medication does then not affect patients' performance, and later ages at seizure onset and surgery do not go hand in hand with better or worse results. Age at testing was (borderline) significant for CYCLE and PPVT. Older patients thus

tended to receive higher (or more age-appropriate) scores than younger patients. Seizure control was found to be significant for CYCLE and Sentence Judgement, with patients whose seizures are controlled consistently outperforming those whose seizures are not controlled. See Table 4 for an overview of all results concerning these variables.

Test Variable	CYCLE	Sentence Judgement	PPVT	Binding subtest of BCP
Seizure Control (One-tailed t-test)	p = ***.028***	p = ***.002***	p = .362	p = .356
Medication (One-tailed t-test)	p = .245	p = .184	p = .400	p = .444
Age at seizure onset (Pearson correlation coefficient, two-tailed t-test)	r = .051 p = .846	r = .204 p = .484	r = .307 p = .215	r = .372 p = 190
Age at surgery (Pearson correlation coefficient, two-tailed t-test)	r = .140 p = .102	r = .443 p = .113	r = .400 p = .100	r = .299 p = .299
Age at testing (Pearson correlation coefficient, two-tailed t-test)	r = .479 p = ***.052***	r = .307 p = .286	r = .497 p = ***.044***	r = -.340 p = .234

Table 4. Results of the statistical analyses for the five remaining clinical variables on the four tests. (Borderline) Significant scores are in bold italics.

6.3. Comparison With Controls

Controls consistently performed better overall than patients, with a higher mean score for all tests and smaller standard deviation for CYCLE and Sentence Judgement. The difference between the control and patient groups is only significant at the .05 level for CYCLE and PPVT, and for Binding only when Sturge-Weber patients are not included. An overview is given in Table 5.

Test	Group	N	Average	S.D.	Two-tailed t-test
CYCLE (DV score)	Control Subjects	14	0.961	0.085	p = ***.011*** (***.009***)
	Hemispherectomy Patients	17 (19)	0.812 (0.820)	0.202 (0.196)	
Sentence Judgement (Raw score /50)	Control Subjects	14	46.07	4.16	p = .240 (.177)
	Hemispherectomy Patients	14 (16)	44.00 (43.81)	4.91 (4.70)	
Binding subtest (Raw score /4)	Control Subjects	14	2.86	0.95	p = ***.047*** (.076)
	Hemispherectomy Patients	14 (16)	2.14 (2.25)	0.864 (0.856)	
PPVT (Standard Score)	Control Subjects	14	114.86	14.85	p < ***.001*** (<***.001***)
	Hemispherectomy Patients	18 (20)	86.94 (87.35)	11.03 (10.99)	

Table 5. Overview of comparison between control subjects and hemispherectomy patients.
Significant scores (at the .05 level) are in bold italics.
Bracketed numbers indicate results of the analysis including Sturge-Weber patients.

7. Discussion

7.1. Major Variables: Side Removed And Aetiology

The primary goal of this study is to determine the predictive values of the variables of side removed and aetiology on the outcome of syntactic ability and lexical knowledge in patients after hemispherectomy. It was hypothesized that only the interaction of the two variables would predict outcomes, at least with respect to syntactic ability. This is shown to be the case in two out of three tests of syntactic ability, with results that are significant at the .10 level. The direction of the trend that became apparent from the interaction was also as hypothesized: although there is no difference in performance between left and right patients with an infarct, such a difference is present between left and right patients with Rasmussen's encephalitis.

The results show that syntactic abilities eventually become lateralised in the left hemisphere. Even after a short period of normal development (as little as one year), syntactic abilities appear to become lateralised to the left. If at this point an insult in the left hemisphere disrupts development, the right hemisphere is already unable to compensate fully for it – though the later the insult happens, the more difficult it becomes for the right hemisphere to compensate. The fact that patients with an acquired aetiology in the left hemisphere consistently outperform those with an

affected right hemisphere is similar to adult stroke patients (similar to what Stark & McGregor 1997 found). However, the results may also suggest that at a very early age the hemispheres may be equipotent. Patients with an early aetiology in the right hemisphere did not appear to be delayed, pointing against left lateralisation before any normal development has taken place.

While these findings relate to syntactic ability, the same results were found for lexical comprehension – though the trend was not as robust. This may be due to the different natures of the two linguistic functions; one being computational and independent, the other non-computational and influenced by IQ and social background. In addition, the typically more distributed nature of this function over the two hemispheres may make the loss of one less detrimental, thus with better recovery abilities in the remaining hemisphere, than a function that is typically strongly lateralised.

7.2. Other Clinical Variables

7.2.1. Seizure Control

The results from the current study also indicate that seizure control has some predictive power. It proved overall significant for the CYCLE and Sentence Judgement tests. The fact that this variable did not prove significant for PPVT may again indicate that syntactic ability and lexical comprehension are supported by different systems. Seizures may then only have an influence on computational functions. Patients whose seizures are controlled outperformed patients whose seizures are not controlled; a trend that held consistently over the four side removed/aetiology groups. Interestingly, the interaction between side removed and aetiology is still visible within the group of five patients whose seizures are not controlled. On all four tests, the right patient with Rasmussen's encephalitis still outperforms the left patients, and the difference between infarct patients is relatively small. Similar results concerning a better outcome when seizures are controlled were found by De Bode (2001) and also Vargha-Khadem et al. (1994) for patients with unilateral lesions. Chiricozzi et al. (2005) on the other hand noted that their patient may have developed so well because of a relatively

mild seizure history, including a seizure free period between CA 6-12, after which seizures deteriorated rapidly (but then were relieved rather quickly again after surgery).

7.2.2. Age At Testing

Age at testing is (almost) significant at the .05 level for CYCLE and PPVT only. Older patients generally got higher or more age-appropriate scores. For CYCLE, this trend may either be due to the few non-syntactic subtests (which may require knowledge that continues to develop throughout life, such as lexical comprehension) included in this test. For PPVT, it is likely to be due to the fact that many older patients have had a longer recovery period after surgery, and thus also a longer time to (re-)learn words (which, unlike syntactic acquisition, is not constrained by maturation at a certain age).

7.3. Presence Of Specific Syntactic Deficits?

Out of the three syntactic tests, the Sentence Judgement task allowed us to examine performance on different syntactic constructions in more detail than just overall ability. Since the Sentence Judgement task is composed out of 12 different sentence types, we looked into the different types further, to detect if patients performed particularly different on any of them. A (near) statistically significant difference was found for three types only: tense ($p = .058$, one-tailed t-test), wh-questions ($p = .005$, one-tailed t-test), and one type of verb sub-categorisation ($p = .024$, one-tailed t-test).

The different performance on tense sentences may be explained by reliance on extra-grammatical knowledge. In case the subject is not certain about the answer by their syntactic ability alone, they may recruit some knowledge of discourse ability. Since tense is currently viewed as being not strictly syntactic (as mentioned above), non-syntactic knowledge can lead to mistakes. Patients accepted more incorrect sentences than control subjects did. This means that they accepted more

sentences where a lexically indicated point in time (for example by using the word *tomorrow*) is not consistent with the tense indicated by agreement on the verb.

Although subjects are indicated to perform differently from controls on wh-questions, there is also one similarity between them: they all perform worse on wh-questions than on any other sentence type. Both control and patient subjects also make more mistakes by rejecting more correct sentences. Where control subjects were having difficulties judging these sentences correctly, patient seemed to find this even more difficult, magnifying their incorrect scores.

The difference in performance on type of subcategorization can also be explained by interference of non-syntactic elements. We have checked the correlation of our results on the syntactic CYCLE test with the lexical PPVT test, and found a high correlation (Pearson's $r = .771$, $p = <.001$, two-tailed t-test). This points toward a common ground between syntax and the lexicon. Subcategorization is marked by features on the verb in the lexicon, to indicate the types of complement that this verb may take in the syntax. It is partly input from the lexicon then that determines the correct use of the verb in the syntax. However, it remains unclear why patients only reject more correct items in one type of subcategorization (namely, determination of the finiteness of the verb's complement), and do not perform differently from controls on the other type (determining whether or not a verb requires obligatory control, $p = .138$, one-tailed t-test).

It is perhaps not surprising that no specific syntactic construction (or their underlying mechanisms) can confidently be pinpointed as areas of impairment. The different aetiologies that cause epileptic seizures are all different in nature, and cause 'damage' to the affected hemisphere in different ways. In addition, not always the same parts of the affected hemisphere are targeted. Still more importantly, none of the aetiologies explicitly target linguistic (or computational) mechanisms in the brain, unlike congenital linguistic impairments such as SLI or dyslexia (Snowling 1995, Leonard 1998, Bishop & Snowling 2004). Rather the deficits observed in hemispherectomy patients are a by-product of a more general disease.

7.4. Aetiologies Revisited

In this study, we have followed the classification of aetiologies by developmental (early) versus acquired (late) aetiologies as described by Curtiss et al. (2001). The results of this study have confirmed this division, but have only included one of each type: infarcts and Rasmussen's encephalitis. Many more aetiologies exist that give rise to intractable epilepsy for which hemispherectomy is considered. The hypotheses set out here predict that they follow the same pattern depending on whether they are developmental or acquired. This is unclear from the literature review, because the numbers are so low: other than patients with an infarct or Rasmussen's encephalitis, it includes only five patients of three different (though all developmental) aetiologies.

A clearer picture may emerge from considering together the results from the literature, the data from the current study (including patients with Sturge-Weber syndrome) and a body of patients reported in current work in progress (Smets & de Bode, manuscript). This body contains data on 34 patients after hemispherectomy, including six different aetiologies (infarct, Rasmussen's encephalitis, cortical dysplasia, hemimegalencephaly, Sturge-Weber syndrome and non-specific diseases). The data for these patients were collected through a questionnaire that was completed by their parents or caregivers, surveying different outcomes. Among other things, general language outcome is reported. Although the questions are not designed to uncover specific deficits in a particular field, they do provide an insight in general recovery and development of the patient.

The data from the three sources is summarised in Table 6, which shows for a new total of 67 patients whether their linguistic skills are good or poor. For the data from the literature review, we assessed outcomes both by patients' scores as well as the authors' analyses (i.e. whether they noted impairments). For our own data, H002 was not included as he did not participate for any of the syntactic tests. Outcome is judged poor if a patient receives a low score on at least two out of the three reliable tests (CYCLE, Sentence Judgement, PPVT). A poor score is indicated by a developmental quotient (= test age divided by chronological age) of 0.666 or less on CYCLE, 40 items (80%) or less

correct on Sentence Judgement and 1.5 S.D. or more below the norm for PPVT. For the data from the questionnaire study (Smets & de Bode, manuscript), Spoken Language Ranks (SLRs, De Bode 2001) were used. This five-point scale indicates a child's general level of linguistic development from 0 (no speech) to 5 (age-appropriate speech); scores below 4 are considered poor.

Side Removed	RIGHT		LEFT		*TOTAL*
Outcome	GOOD	BAD	GOOD	BAD	
Literature Review	2 RE			3 RE	*5 RE*
	1 inf		2 inf		*3 inf*
			2 S-W		*2 S-W*
		2 HME			*2 HME*
		1 cyst			*1 cyst*
			1 NS		*1 NS*
Current Study	4 RE			3 RE	*7 RE*
	1 inf	1 inf	6 inf	2 inf	*10 inf*
		1 S-W	1 S-W		*2 S-W*
Questionnaire Study	4 RE		1 RE (early)	1 RE	*6 RE*
	2 inf		2 inf		*4 inf*
		1 S-W	2 (0) S-W		*3 (1) S-W*
	6 NS		3 NS	1 NS	*10 NS*
		2 (1) CD			*2 (1) CD*
		4 (3) HME	3 HME	2 (1) HME	*9 (7) HME*
Total	**10 RE**		**1 RE (early)**	**7 RE**	***18 RE***
	4 inf	**1 inf**	**10 inf**	**2 inf**	***17 inf***
	6 NS		**4 NS**	**1 NS**	***11 NS***
		6 (5) HME	**3 HME**	**2 (1) HME**	***11 (9) HME***
		2 S-W	**5 (3) S-W**		***7 (5) S-W***
		2 (1) CD			***2 (1) CD***
		1 cyst			***1 cyst***

Table 6. Results for 67 patients using data from the literature, the current study and a questionnaire study.
Numbers in brackets exclude those who are younger than CA 6 years.
RE = Rasmussen's encephalitis
inf = infarct
S-W = Sturge-Weber syndrome
HME = hemimegalencephaly
CD = cortical dysplasia
NS = non-specific disease

The eighteen patients with Rasmussen's encephalitis (the only acquired aetiology in the sample) continue to show the predicted distribution, with the exception of one left patient. However, the onset of aetiology for this patient was very early at just 2 months old, which may explain the good recovery. The seventeen patients with an infarct also still follow the same trend; they generally perform well, with the exception of a small proportion whose seizures are not controlled or whose scores fall just below the cut-off scores (all are from the current study). With respect to the remaining aetiologies, the clearest outcomes are obtained by the group of patients whose aetiology is not entirely certain (non-specific diseases), with ten out of eleven patients performing well. Since the aetiologies are not precisely identified, we cannot make any assumptions as to their natures and consequences for outcomes. Within the group of patients with hemimegalencephaly, the outcomes are only uniform for patients with an affected right hemisphere – they all perform poorly, going against the prediction. For patients with an affected left hemisphere, on the other hand, outcome does not seem predictable. But both patients with hemimegalencephaly in the left hemisphere perform poorly and have not had seizures controlled by surgery, so this factor may again play a role here. Though the numbers are low, the outcomes for patients with Sturge-Weber syndrome seem uniform, though going precisely against what we expect: all right patients perform poorly, and all left patients do well. Numbers are still lower for patients with cortical dysplasia and a cyst, but they perform similarly to patients with Sturge-Weber syndrome with poor scores if the right hemisphere is involved.

Thus, the findings for Rasmussen's encephalitis and infarcts are confirmed, but the results for all other aetiologies do not follow the predicted trend. Specifically patients with Sturge-Weber syndrome and hemimegalencephaly show differences between right and left patients where these are not predicted due to their developmental (early) nature. Moreover, the difference goes against what would be predicted taking into account linguistic lateralization. If anything, it should be patients after left hemispherectomy who are more impaired than the right patients. Seizure control cannot

explain these findings per se, because all of these right patients have seizures controlled at the time of testing (though seizure histories are unknown). Even if no data is available to compare the right patients with cortical dysplasia or a cyst to left patients, the right patients should also perform well.

These data show that not all aetiologies classified here as developmental show the same trends with respect to left and right patients. As already suggested by Curtiss & de Bode (1999), perhaps 'developmental' is more of an umbrella term containing many aetiologies that may be too different to be classified as one type, and can be broken down into smaller categories. If we term what we have so far called 'developmental' simply as 'early' or 'pre-/perinatal' (and 'acquired' as 'late' or 'postnatal'), the different aetiologies may be further divided into acquired, progressive and developmental types (Liégeois et al. 2008b). Infarcts would then fall in the acquired type (Jonas et al. 2004); although its consequences continue in the long term giving it developmental characteristics, its onset is rather sudden. Hemimegalencephaly and cortical dysplasia are developmental aetiologies (Jonas et al. 2004), as their cause lies in abnormal neuronal development. Sturge-Weber syndrome has been classified as a progressive aetiology (Maria et al. 1998). (For the sake of completeness, the postnatal aetiology Rasmussen's encephalitis is also progressive [Vining et al. 1993]). This more detailed classification seems to reflect the differences with regard to outcomes better, with on the one hand acquired aetiologies showing good recovery rates for both sides, and progressive and developmental aetiologies showing a difference between right and left patients. The remaining hemisphere that is able to recover better then depends on whether the onset is early or late.

7.5. For Or Against Equipotentiality: A Middle Ground

Regarding the reason why early acquired aetiologies may perform differently from developmental and progressive aetiologies, we first look into the equipotentiality debate again. Recently, a third view has emerged that forms a compromise between the two main views. Although the left hemisphere does contain biases to accommodate syntactic functions, the right hemisphere is also able to support linguistic development if the left is damaged at an early age (Bates & Roe 2001).

In addition, the right hemisphere is attributed a larger role in initial stages of acquisition than was previously believed (Locke 1993, 1997, Bates & Roe 2001).

Locke (1997) in particular has strongly advocated this idea. He argues that all linguistic development up until the age of around two years takes place in the right hemisphere. The earliest development is concerned with social aspects of language (Papousek & Papousek 1989) and the detection of prosodic patterns (Mehler et al. 1988), which allows the child to get familiarised with the language he or she hears. From around 7 months old, infants start to develop lexical comprehension in a holistic manner, without distinguishing between discrete lexical items within a phrase (Echols 1993). Only when syntactic development starts around age two, does the left hemisphere progressively take over. Following these ideas, Locke (1997) specifically predicts that early right hemisphere damage (or onset of aetiology which effectively behaves like damage) will then lead to deficits in lexical comprehension, but not in syntactic development.

However, the patients who do not perform well in Liégeois (2008a, 2008b) did so across the board as mentioned above and not just on lexical comprehension. This clearly goes against Locke's (1997) predictions. Still, if we consider his theory in the light of a more modern architecture of the lexicon, these results may be explained. In the Minimalist framework of the generative tradition (Chomsky 1995), an entry in the lexicon is no more than a collection of features relating to different aspects of the word in question (phonological, semantic, syntactic, etc.). The syntactic module is concerned with the manipulation of such syntactic features. Therefore, if lexical acquisition is impaired and, as a result, features are not appropriately acquired, there may be consequences for syntactic acquisition, which then cannot make use of these features appropriately. An apparent impairment in the acquisition of syntax may be the result, pulled by an impairment in the lexicon.

The difference between early acquired aetiologies on the one hand, and developmental and progressive (non-acquired) aetiologies on the other hand, lies in the type of onset. In an acquired aetiology like an infarct, the onset is very sudden and the damage is considerable very soon. In effect,

the affected hemisphere is almost immediately entirely wiped out (De Bode 2001). In contrast, non-acquired aetiologies do not have such an abrupt onset but gradually become more severe over time, without any immediate large scale damage (Jonas et al. 2004). The chances for successful compensation by the other hemisphere are different in the two cases. In the case of acquired aetiologies, the affected hemisphere is almost completely unable to support its original functions and takeover by the healthy hemisphere is easier (not unlike the 'larger lesions are better' hypothesis, Irle [1987], Kertesz [1988]). In the absence of extensive damage in non-acquired aetiologies, the affected hemisphere may attempt to carry out its functions as far as it is still able to, disfavouring takeover by the healthy hemisphere.

Therefore, at the onset of an early acquired aetiology, transfer of functions is often successful in either hemisphere. When the right hemisphere is affected, early lexical comprehension can proceed normally in the left hemisphere, and syntactic development may be supported by the right hemisphere in the reverse situation. But when an early non-acquired aetiology sets on, the side of injury will be crucial to the eventual outcome. If the left hemisphere is affected, lexical comprehension may still develop normally in the right hemisphere. By the time syntactic development is set to begin, the damage in the left hemisphere may have become considerably more severe – perhaps enough for the right hemisphere to take over. If so, both lexical and syntactic development will reach good levels. But if the right hemisphere is affected by a non-acquired aetiology, it may try to support lexical comprehension because of small initial damage, and lead to deficits in this linguistic domain. Even if the left hemisphere is unaffected, syntactic development then does not proceed normally, because it cannot make appropriate use of the features it requires. As a result, linguistic impairments are noted across the board with poor performance on both lexically and syntactically oriented tasks. Indeed, this is precisely what was found in the literature, the current study and the questionnaire study (Smets & de Bode, manuscript).

8. Future Research

The current research has not been able to investigate the non-acquired aetiologies identified above. Therefore, further research is necessary to identify if patients with early developmental and progressive aetiologies indeed perform similarly, and may be classified together as non-acquired. If so, this research should also investigate whether patients with an acquired aetiology perform differently from patients with a non-acquired aetiology, and how such a difference may be characterised. As in the present study, not all patients are hypothesized to perform equally depending on whether their left or right hemisphere is removed. Both the variables of side removed and aetiology are thus again the major variables involved.

The key to the success of this research is the inclusion of a large number of patients – something that poses a problem to many studies in the field. The aetiologies giving rise to intractable epilepsy in a single hemisphere are all rare, and the number of patients who have undergone hemispherectomy is even lower. Moreover, patients need to be old enough to have developed some syntactic ability; considering their general delay as compared to neurologically normal children, they should be at least six years of age. To ensure that they have completely recovered from their surgery, testing should take place at least three years after surgery.

Ideally, around 50 patients of each aetiology would be included to enhance statistical power, divided equally over right and left patients after hemispherectomy. More realistically, this number lies around a maximum of 15-20 patients, and is not likely to include equal numbers of right and left patients. The problem intensifies if only patients are included whose seizures are controlled, to avoid possible confounds. However, since it is known that seizure control may play a role regarding outcome, this may be taken into account in the data analysis by including it as a variable. The results of patients without seizure control are still valuable, because they may still reflect same the trends as patients with seizure control, only with reduced scores (as in the current study). But even with some

lower numbers of patients, some trends may still be found, even if they are not statistically significant at the strict .05 level.

The difference between left and right patients within a non-acquired aetiology is predicted to be apparent in both lexical comprehension and syntactic ability, as described above. Therefore, tests for both of these functions should be used. We propose the use the most recent versions of the TROG (TROG-2, Bishop 2003) and PPVT (PPVT-III, Dunn & Dunn 1997). These are two standardised tests that have several advantages: they are up-to-date, have been age-normed and are widely used, which makes it easier to directly compare different studies using them.

The results of this research will clarify the outcome of linguistic abilities for patients after hemispherectomy with an early aetiology. If the interaction between aetiology and side removed proves to be significant, then we have found a basis from predicting outcomes for both early and late aetiologies (though the trend will be different depending on the precise aetiologies). In addition, it provides indirect evidence for the theoretical framework arguing for early linguistic development in the right hemisphere.

9. Conclusion

The literature has not been conclusive on the outcomes of syntactic ability in patients after hemispherectomy. It does however suggest that both factors of side removed and aetiology need to be taken into account together. Our study partly overcomes the methodological shortcomings of the previous studies, and our results confirm this hypothesis. Namely, patients with an infarct – an aetiology that was initially classified as developmental – did not show a difference in performance depending on whether their right or left hemisphere was removed. However, patients with Rasmussen's encephalitis – an aetiology initially classified as acquired – did show a difference in performance, with patients whose right hemisphere was removed consistently outperforming those whose left hemisphere was removed. In addition, the variables of seizure control and age at testing

were found to influence performance. These results show that the new born's brain may be equipotent at first, but that this may be erased as soon as normal linguistic development takes place, when syntactic abilities become lateralised to the left hemisphere.

This study has looked into two aetiologies, but many more exist for which hemispherectomy may be performed, in particular with a pre/perinatal onset. A closer examination of the small amount of data available on these aetiologies suggests that they may be too different to all be contained under the 'developmental' term, but are rather separate aetiology types that all have an early onset (as opposed to those with a late onset). It also suggests that patients of one of these early types (infarct) perform differently from the other two types (developmental and progressive), who seem to perform alike. That is, the latter two groups show a difference between right and left patients, with left patients outperforming right patients. This would fit in a more recent view on equipotentiality of the new born brain, which takes into account a larger role for the right hemisphere in early language acquisition. Again the side removed and aetiology variables appear to be of importance here, but further research is necessary to confirm these propositions.

References

Adams, C.B.T. (1983) Hemispherectomy – A Modification. In: *Journal of Neurology, Neurosurgery and Psychiatry*, Vol. 46, pp. 617-619

Aram, D. M., Ekelman, B., Rose, D., and Whitaker, H. (1985) Verbal And Cognitive Sequelae Following Unilateral Lesions Acquired In Early Childhood. In: *Journal Of Clinical And Experimental Neuropsychology,* Vol. 7, pp. 55-78.

Aram, D., Ekelman, B. and Whitaker, H. (1986) Spoken Syntax In Children With Acquired Unilateral Hemisphere Lesions. In: *Brain And Language*, Vol. 27, pp. 75-100.

Ballantyne, A. O., Scarvie, K. M., and Trauner, D. A. (1994). Verbal And Performance IQ Patterns In Children After Perinatal Stroke. In: *Developmental Neuropsychology,* Vol. 10, pp. 39-50.

Bates, E., Thal, D., Trauner, D., Fenson, J., Aram, D., Eisele, J. and Nass, R. (1997) From First Words To Grammar In Children With Focal Brain Injury. In: *Developmental Neuropsychology*, Vol. 13, No. 3, pp. 275-343

Bates, E., Reilly, J., Wulfeck, B., Dronkers, N., Opie, M., Fenson, J., Kriz, S., Jeffries, R., Miller, L-R. and Herbst, K. (2001) Differential Effects Of Unilateral Lesions On Language Production In Children And Adults. In: *Brain And Language*, Vol. 79, pp. 223-265

Bates, E. and Roe, K. (2001) Language Development In Children With Unilateral Brain Injury. Chapter 20 in: Nelson, C.A. and Luciana, M. (eds.), *Handbook Of Developmental Cognitive Neuroscience.* Cambridge, MA: MIT Press. pp. 281-308

Binding, Control and Presupposition (BCP). [Unpublished test. University of California, Los Angeles]

Bishop, D.V.M. (1982) *TROG: Test For Reception Of Grammar*. Manchester: Chapel Press

Bishop, D.V.M. and Snowling, M.J. (2004) Developmental Dyslexia and Specific Language Impairment: Same or Different? In: *Psychological Bulletin*, Vol. 130, No. 6, pp. 858–886

Brown, R. (1973) *A First Language: The Early Stages*. Cambridge, MA: Harvard University Press

Carson, B.S., Javedan, S.P., Freeman, J.M., Vining, E.P.G., Zuckerberg, A.L., Lauer, J.A. and Guarnieri, M. (1996) Hemispherectomy: A Hemidecortication Approach And Review of 52 Cases. In: *Journal Of Neurosurgery*, Vol. 84, pp. 903-911

Chiricozzi, F., Chieffo, D., Battaglia, D., Iuvone, L., Acquafondata, C., Cesarini, L., Sacco, A., Chiera, R., Di Rocco, C. and Guzzetta, F. (2005) Developmental Plasticity After Right Hemispherectomy In An Epileptic Adolescent With Early Brain Injury. In: *Child's Nervous System*, Vol. 21, 960-969

Chomsky, C. (1969) *The Acquisition Of Syntax In Children From 5 To 10*. Cambridge, MA: MIT Press

Chomsky, N. (1981) *Lectures On Government And Binding*. Dordrecht: Foris

Chomsky, N. (1995) *The Minimalist Program*. Cambridge, MA: MIT Press

Curtiss, S. and De Bode, S. (1998) Linguistics Outcomes For Hemispherectomised Children. In: Greenhill, A., Hughes, M., Littlefield, H. and Walsh, H. (eds.) *BUCLD 22: Proceedings Of The 22nd Annual Boston University Conference On Language Development*. Somerville, MA: Cascadilla Press

Curtiss, S. and De Bode, S. (1999) Age And Etiology As Predictors Of Language Outcome Following Hemispherectomy. In: *Developmental Neuroscience*, Vol. 21, pp. 174–181

Curtiss, S., De Bode, S. and Mathern, G. (2001) Spoken Language Outcome After Hemispherectomy: Factoring In Etiology. In: *Brain And Language*, Vol. 79, pp. 379-396

Curtiss, S. and Yamada, J. (1988) *Curtiss-Yamada Comprehensive Language Evaluation (CYCLE)*. University of California, Los Angeles

De Bode (2001) *Language In The Isolated Right Hemisphere: Predicting And Quantifying language Outcomes Following Hemispherectomy*. Unpublished PhD Dissertation, University of California, Los Angeles

Dennis, M. (1980) Capacity And Strategy For Syntactic Comprehension After Left Or Right Hemidecortication. In: *Brain And Language*, Vol. 10, pp. 287-317

Dennis, M. and Kohn, B. (1975) Comprehension Of Syntax In Infantile Hemiplegics After Cerebral Hemidecortication: Left-Hemisphere Superiority. In: *Brain And Language*, Vol. 2, pp. 472-482

Dennis, M. and Kohn, B. (1985) The Active-Passive Test: An Age Referenced Clinical Test Of Syntactic Discrimination. In: *Developmental Neuropsychology*, Vol. 1, No. 2, pp. 113-137

Dennis, M. and Whitaker, H.A. (1976) Language Acquisition Following Hemidecortication: Linguistic Superiority Of The Left Over The Right Hemisphere. In: *Brain And Language*, Vol. 3, pp. 404-433

Duchowny, M. (1993) Identification Of Surgical Candidates And Timing Of Operation: An Overview. In: Wyllie, E. (ed.) *The Treatment Of Epilepsy: Principles And Practices*. Philadelphia, PA: Lea and Febiger. pp. 999-1008

Dudley, J. and Delage, J. (1980) *Tests De Langage Dudley-Delage (TLDD)*. St-Lambert: Les Editions De l'ABC

Dunn, L.M. and Dunn, L.M. (1997) *Peabody Picture Vocabulary Test – Third Edition (PPVT-III).* Circle Pines, MN: AGS

Echols, C. H. (1993) A Perceptually-Based Model Of Children's Earliest Productions. In: *Cognition,* Vol. 46, pp. 245–296.

Feldman, H.M., Holland, A.L., Kemp, S.S. and Janosky, J. (1992) Language Development After Unilateral Brain Injury. In: *Brain And Language*, Vol. 42, pp. 89-102

Feldman, H.M. (1994) Language Development After Early Unilateral Brain Injury: A Replication Study. Chapter 4 in: Tager-Flusberg, H. (ed.) *Constraints On Language Acquisition: Studies On Atypical Children*. Hillsdale, NJ: Lawrence Erlbaum Associates. pp. 75-90

Irle, E. (1987) Lesion Size And Recovery Of Function: Some New Perspectives. In: *Brain Research Reviews*, Vol. 12, No. 3, pp. 307-320

Jonas, R., Nguyen, S., Hu, B., Asarnow, R.F., LoPresti, C., Curtiss, S., de Bode, S., Yudovin, S., Shields, W.D., Vinters, H.V., and Mathern, G.W. (2004) Cerebral Hemispherectomy: Hospital Course, Seizure, Developmental, Language, And Motor Outcomes. In: *Neurology*, Vol. 62, pp. 1712-1721

Kempler, D. (1986) The Kempler Comprehension Task. [Unpublished]

Kertesz, A. (1988) What Do We Learn From Recovery From Aphasia? In: *Advances In Neurology*, Vol. 47, pp. 277-292

Lee, L. (1969) *Northwestern Syntax Screening Test (NSST)*. Evanston,IL: Northwestern University Press

Lenneberg, E. (1967) *Biological Foundations Of Language*. New York, NY: John Wiley & Sons

Leonard, L. B. (1998). *Children With Specific Language Impairment*. Cambridge, MA: MIT Press.

Liégeois, F., Connelly, A.L., Baldeweg, T., Vargha-Khadem, F. (2008a) Speaking With A Single Cerebral Hemisphere: fMRI Language Organisation After Hemispherectomy In Childhood. In: *Brain And Language*, Vol. 106, pp. 195-203

Liégeois, F., Cross, J.H., Polkey, C., Harkness, W. and Vargha-Khadem, F. (2008b) Language After Hemispherectomy In Childhood: Contributions From Memory And Intelligence. In: *Neuropsychologia*, Vol. 46, pp. 3101-3107

Locke, J.L. (1993) The Role Of The Face In Vocal Learning And The Development Of Spoken Language. In: De Boysson-Bardies, B., De Schonen, S., Jusczyk, P., MacNeilage, P. and Morton, J. (eds*.) Developmental Neurocognition: Speech And Face Processing In The First Year Of Life*. Dordrecht: Kluwer

Locke, J.L. (1997) A Theory Of Neurolinguistic Development. In: *Brain And Language*, Vol. 58, pp. 265-326

Maria, B.L., Neufeld, J.A. and Rosainz, L.C. (1998) Central Nervous System Structure And Function In Sturge-Weber Syndrome: Evidence Of Neurologic And Radiologic Progression. In: *Journal Of Child Neurology*, Vol. 13, No 12, pp. 606-18

Mariotti, P., Iuvone, L., Torrioli, M.G. and Silveri, M.C. (1998) Linguistic And Non-Linguistic Abilities In A Patient With Early Left Hemispherectomy. In: *Neuropsychologia*, Vol. 36, No. 12, pp. 1303-1312

Martínez-González, J.A., Gupta, A., Kotagal, P., Lachhwani, D., Wyllie, E., Lüders, H.O. and Bingaman, W.E. (2005) Hemispherectomy For Catastrophic Epilepsy In Infants. In: *Epilepsia*, Vol. 46, No. 9, pp. 1518-1525

Mathern, G.W., Giza, C.C., Yudovin, S., Vinters, H.V., Peacock, W.J., Shewmon, D.A. and Shields, W.D. (1999) Postoperative Seizure Control And Antiepileptic Drug Use In Pediatric Epilepsy Surgery Patients: The UCLA Experience, 1986-1997. In: *Epilepsia*, Vol. 40, No. 12, pp. 1740-1749

Mehler, J., Jusczyk, P., Lambertz, G., Halsted, N., Bertoncini, J. and Amiel-Tison, C. (1988) A Precursor Of Language Acquisition In Young Infants. In: *Cognition*, Vol. 29, pp. 143-178

Ménard, A., Le Normand, M-T., Rigoard, M-T. and Cohen, H. (2000) Language Development In A Child With Left Hemispherectomy. In: *Brain And Cognition*, Vol. 43, pp. 332-340

Miceli, G., Laudanna, A., Burani, C. and Capasson, R. (1994) *Batteria Per l'Analisi Dei Deficit Afsici (B.A.D.A.)*, Universita Cattolica Del Sacro Cuore, Rome

Molfese, D. and Segalowitz, S. (1988) *Brain Lateralization In Children*. New York: The Guildford Press

Papousek, M. and Papousek, H. (1989) Forms And Functions Of Vocal Matching In Interactions Between Mothers And Their Precanonical Infants. In: *First Language*, Vol. 9, pp. 137-158

Peacock, W.J. (1995) Hemispherectomy For The Treatment Of Intractable Seizures In Childhood. In: *Neurosurgery Clinics Of North America*, Vol. 6, pp. 549-563

Pinker, S. (1994) *The Language Instinct*. London: Penguin Books

Pizzamiglio, L. and Parisi, D. (1970) Studies On Verbal Comprehension In Aphasia. In: Flores d'Arcais, G.B. and Levelt, W.J.M. (eds.) *Advances In Psycholinguistics*. Amsterdam: North Holland

Pulsifer, M.B., Brandt, J., Salorio, C.F., Vining, E.P.G., Carson, B.S. and Freeman, J.M. (2004) The Cognitive Outcome Of Hemispherectomy In 71 Children. In: *Epilepsia*, Vol. 45, No. 3, pp. 243-254

Rankin, J.M., Aram, D.M. and Horwitz, S.J. (1981) Language Ability In Right And Left Hemiplegic Children. In: *Brain And Language*, Vol. 14, pp. 292-306

Rasmussen, T. and Milner, B. (1977) The Role Of Early Left-Brain Injury In Determining Lateralization Of Cerebral Speech Functions. In: *Annals Of The New York Academy Of Sciences*, Vol. 299, pp. 355-369

Reilly, J.S., Bates, E. and Marchman, V.A. (1998) Narrative Discourse In Children With Early Focal Brain Injury. In: *Brain And Language*, Vol. 61, pp. 335-375

Riva, D. and Cazzaniga, L. (1986) Late effects Of Unilateral Brain Lesions Sustained Before And After Age One. In: *Neuropsychologia*, Vol. 24, No. 3, pp. 423-428

Schwartz, M.F., Saffran, E.M. and Marin, O.M.S. (1980) The Word Order Problem In Agrammatism: I. Comprehension. In: *Brain And Language,* Vol. 10, pp. 249-262

Semel, E., Wiig, E.H. and Secord, W. (2000) *Clinical Evaluation Of Language Fundamentals-Receptive (CELF-R) – Third Edition UK*. Oxford: Hartcourt Assessment

Sentence Judgement. [Unpublished test. University of California, Los Angeles]

Smets, L.M.K. and De Bode, S. (manuscript) Language Outcome After Hemispherectomy: A Questionnaire Study [Unpublished]

Snowling, M. (1995) Phonological Processing And Developmental Dyslexia. In: *Journal Of Research In Reading*, Vol. 13, No. 2, pp. 132-138

Stark, R.E., Bleile, K., Brandt, J., Freeman, J. and Vining, E.P.G. (1995) Speech-Language Outcomes Of Hemispherectomy In Children And Young Adults. In: *Brain And Language*, Vol. 51, pp. 406-421

Stark, R.E. and McGregor, K.K. (1997) Follow-Up Study Of A Right- And A Left-Hemispherectomized Child: Implications For Localization And Impairment Of Language In Children. In: *Brain And Language*, Vol. 60, pp. 222-242

Telfeian, A.E., Berqvist, C., Danielak, C., Simon, S.L. and Duhaime, A-C. (2002) Recovery Of Language After Left Hemispherectomy In A Sixteen-Year-Old Girl With Late-Onset Seizures. In: *Pediatric Neurosurgery*, Vol. 37, pp. 19-21

Trudeau, N., Colozzo, P., Sylvestre, V. and Ska, B. (2003) Language Following Functional Left Hemispherectomy In A Bilingual Teenager. In: *Brain And Cognition*, Vol. 53, pp. 384-388

Van Lancker, D. (2001) Forum: Is Your Syntactic Component Really Necessary? In: *Aphasiology*, Vol. 15, No. 4, pp. 343-406

Vanlancker-Sidtis, D. (2004) When Only The Right Hemisphere Is Left: Studies In Language And Communication. In: *Brain And Language*, Vol. 91, pp. 199-211

Vargha-Khadem, F., Isaacs, E. and Muter, V. (1994) A Review Of Cognitive Outcome After Unilateral Lesions Sustained During Childhood. In: *Journal Of Child Neurology,* Vol. 9, Supplement No. 2, pp. 2S67-2S73

Vargha-Khadem, F., Carr, L.J., Isaacs, E., Brett, A., Adams, C. and Mishkin, M. (1997) Onset Of Speech After Left Hemispherectomy In A Nine-Year-Old Boy. In: *Brain*, Vol. 120, pp. 159-182

Vicari, S., Albertoni, A., Chilosi, A.M., Cipriani, P., Cioni, G. and Bates, E. (2000) Plasticity And Reorganization During Language Development In Children With Early Brain Injury. In: *Cortex*, Vol. 36, pp. 31-46

Vining, E.P.G., Freeman, J.M., Brandt, J., Carson, B.S. and Uematsu, S. (1993) Progressive Unilateral Encephalopathy Of Childhood (Rasmussen's Syndrome): A Reappraisal. In: *Epilepsia*, Vol. 34, No 4, pp. 639-650

Witelson, S. (1987) Neurobiological Aspects Of Language In Children. In: *Child Development*, Vol. 58, pp. 653-688

APPENDIX A

Overview of the literature review.

Article	Number of patients	Patient nr	R/L	Aetiology	Age at onset seizures	Surgery age	Age at test	Test used
Stark & McGregor (1997)	2	1	R	Rasmussen's encephalitis	2;0	5;8	surgery- 9;6	CYCLE, TROG
		2	L	Rasmussen's encephalitis	1;6	4;0	surgery- 9;6	
Varga-Khadem et al. (1997)	1	3	L	Sturge-Weber syndrome	6 days	8;6	8;2-14;11	TROG
Mariotti et al. (1998)	1	6	L	Sturge-Weber syndrome	0;3	3	20	B.A.D.A, Pizzamiglio & Parisi 1970, Schwartz 1980
Trudeau et al. (2003)	1	4	L	Rasmussen's encephalitis	5	16	16;5	TLDD
Chiricozzi et al. (2005)	1	5	R	Infarct	4;6	14;10	19;6	B.A.D.A
Vanlancker-Sidtis (2004)	1	N/A	L	N/A	0;5	5;6	49;6	Kempler Comprehension, Active-Passive
Liégeois et al. (2008)	6	7	L	Rasmussen's encephalitis	3;6	4;1	12;5	CELF-R
		8	R	Rasmussen's encephalitis	6;0	11;9	22;10	
		9	L	Infarct	6;0	11;11	13;6	
		10	L	Infarct	0;1	9;11	14;11	
		11	R	Hemimegalencephaly	0;7	4;2	12;5	
		12	R	Cyst	4;6	11;11	21;2	
Liégeois et al. (2008b)	30	(13)	R = 13 L = 17	Cortical dysplasia = 1 Sturge-Weber syndrome = 3 Infarct = 12 Rasmussen's encephalitis = 8 Other=6	0;1-10;0	0;4-15;7	7;11-24;3	CELF-R

APPENDIX B

Syntactic test evaluation

One disadvantage of many (often older) syntactic tests is that they use test items where pronouns or prepositions are the focus; for example whether the patient knows that *he* should refer to a male person, or *on* to the top of an object. However, is not at all clear that pronouns and prepositions are in fact part of the morpho-syntactic system – instead, in recent generative frameworks, they belong to the semantics of an utterance (Chomsky 1998[9], Hornstein et al. 2005[10]). Pronouns select a person from the context and prepositions indicate a direction or location.

Test for the Reception Of Grammar (TROG), Bishop (1982)

The TROG test, a sentence-picture matching task, seems to be a relatively good measure of syntactic ability. The items really seem to test (morpho-)syntactic knowledge, and this is mostly achieved by selecting appropriate distractors to each item. In the example of an easy item (Figure 1) testing pronoun number marking, the grammatical distractor changes subject number, and so there is different verb agreement. Moreover, there is not only a grammatical distractor but also two lexical distracters which can indicate a non-syntactic deficit if chosen by the patient. One disadvantage is the treatment of pronouns and prepositions as syntactic elements. In addition, the test is normed for all ages, although syntactic development is generally accepted to reach an adult level around age 9. A test score of over 9 years, but below one's CA would then falsely indicate a syntactic impairment, more likely caused by the more semantic items instead.

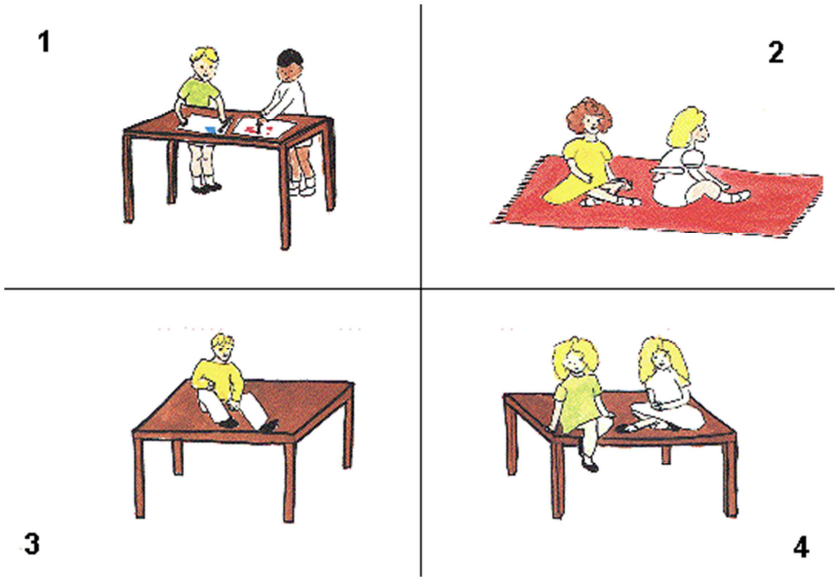


Figure 1. "They are sitting on the table".

Syntactic comprehension from Tests de Langage Dudley-Delage (Dudley & Delage 1980)

The syntactic comprehension subpart of the Tests de Langage Dudley-Delage (TLDD) is in fact an adapted French version of the Northwestern Syntax Screening Test (NSST) developed by Lee (1969). However, it is not clear whether this test has only been translated into French, or whether other substantial changes have been made. The TLDD syntactic comprehension subpart reported by Trudeau et al (2003) is comprised of 48 sentence-picture matching items, yet the receptive portion of the NSST only has 20 items. It remains unclear where the other items come from, or what they focus on. Another important disadvantage is that the examiner

[9] Chomsky, N. (1998) *The Minimalist Program*. Cambridge, MA: MIT Press
[10] Hornstein, N., Nunes, J. and Grohmann, K.K. (2005) *Understanding Minimalism*. Cambridge, UK: CUP

describes two of the four pictures first, then asks the patient to point at one of these two. An example is shown in (1) below. It seems a little odd to describe only two of the four pictures, which may confuse the patient, possibly leading him or her to give an incorrect answer which is not due to syntactic ability. It seems more felicitous to either describe all four pictures or preferably none, since the description and especially the command at the end are syntactically substantially more complex than the target sentence. The NSST also treats at least prepositions (but possibly also pronouns) as part of morphosyntax.

(1) Examiner: "On one of these pictures the cat is behind the chair; on another, the cat is under the chair. Now show me the one in which the cat is behind the chair."

B.A.D.A. – morphosyntactic comprehension (Miceli et al. 1994)

The morphosyntactic subcomponent of the B.A.D.A. battery is also a sentence-picture matching test. There are two ways of presenting the target sentence: either auditorily or visually. Prepositions and pronouns do not seem to be tested, but Mariotti et al. (1998) do note that the target and distractor may differ in only semantic features. The reasoning behind this is that these features are (in part) necessary to identify which elements are subjects, objects or predicates. Still this does mean that in these cases, possibly not syntactic ability but semantic notions are being tested. It is unclear from the data available whether this really poses a problem.

Figure 2. Example: "Il bambino salute le bambine"
The boy greets the children
"The boy greets the children."

Sentence comprehension by Pizzamiglio & Parisi (1970)

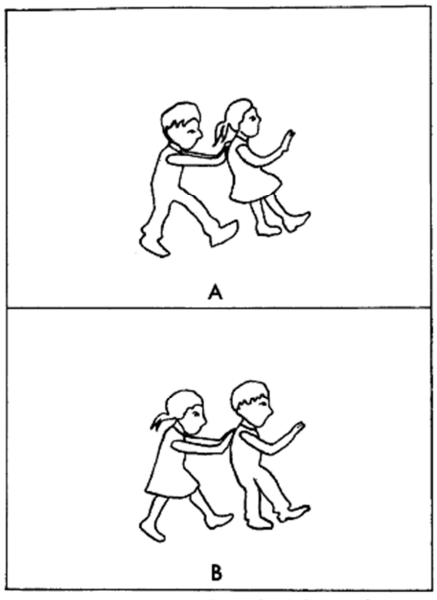
This sentence comprehension task is another sentence-picture matching test, with three types of contrast involved. First of all, there is a contrast in what Pizzamiglio & Parisi (1970) call grammatical units. They do not define this term, but it is likely that they mean separate morphemes such as prepositions. It is unclear whether

they treat pronouns the same way. Secondly, bound morphemes are a contrast tested. This seems a more genuine testing of syntactic ability, because it requires the analysis of agreement (not only on verbs but also subject and objects).The third type of contrast is defined as "other properties of words in the sentence". This very vague description mostly seems to refer to reversible sentences, as one of their examples given indicates. Although this does indeed test word order (and because this is relevant in the syntax of Italian, also syntactic ability), it is unclear if there are other types of items included here. Altogether, because of the rather vague descriptions and the inclusion of prepositions as grammatical units, this test is probably the weakest of all tests reviewed here.

Active-Passive Test (Dennis & Kohn 1985)

The Active-Passive test is also a test based on sentence-picture matching. There are only four types of possible target sentences: active affirmative, active negative, passive affirmative and passive negative. However, in this test it is not clear that negative morphemes are really syntactic, or whether they are more semantic like prepositions and pronouns. This problem is even brought up by one of the hemispherectomy studies (Vanlancker-Sidtis 2004) and is one that has not been resolved yet. It seems that indeed this debate is not an easy one to solve, because negativity does not seem purely syntactic but also not purely semantic. Furthermore, theories of generative syntax can diverge greatly in their treatment of negative morphemes, specifically whether they should be considered as a category of their own (as is generally the case in Minimalism) or whether they can be included in a more general category (adverbs, in many older frameworks) (see Hornstein et al. 2005:194[11] for a short exposition).

Figure 3. Example: *"The girl pushes the boy."*



Test of comprehension of reversible active and passive sentences (Schwartz et al. 1980)

This is another sentence-picture matching test, with only active sentences and passive sentences (where thematic roles have been inversed). The sentences are all reversible, so that subjects cannot guess from the context which argument is the agent and which is the patient, and both conditions are included. A second

[11] Hornstein, N., Nunes, J. and Grohmann, K.K. (2005) *Understanding Minimalism*. Cambridge, UK: CUP

advantage is that no negative sentences are used, which could have complicated the sentences in a non-syntactic way (as above).

Active example sentence: *"The clown applauds the dancer."*
Passive example sentence: *"The clown is applauded by the dancer."*

CYCLE (Curtiss & Yamada 1988)
The receptive portion of the CYCLE battery is mostly composed of sentence-picture matching tests though some involve executing short commands. The tests are not all syntactic in nature, but those that are marked as being syntactic, conform to the recent standards that elements that are not involved in agreement or movement, are not syntactic. Prepositions and pronouns are regarded as being semantic in nature, and tests for elements that are not clearly one or the other (for example, pronouns marked for case) are labeled as both syntactic and semantic so that the researcher may decide which option to choose.

Example syntactic test item: "*The girl is pushing the boy*."

Kempler Comprehension Task (Kempler 1986)
Not much information is available on this unpublished test, which is another sentence-picture matching task. It is described by Vanlancker-Sidtis (2004) as testing "verbal expressions of increasing syntactic complexity, which include passive voice and embedded clauses" (2004:201). These do indeed classify as syntactic constructions, but it remains unclear how this test deals with the more controversial elements.

CELF-R (Semel et al. 2000)
The receptive half of the CELF much resembles that of CYCLE. Since it is a tool for assessment of general receptive language, it does not only include tests of syntactic ability but also tests that are clearly semantic in nature. One advantage of the test is that prepositions are not regarded as syntactic elements.

Example from a syntactic picture selection subtest: *"The boy is being followed by the dog."*
Example from a semantic subtest (choose two that match): *"simple, happy, easy"* (simple, easy)