Brain & Language 140 (2015) 35–48

Contents lists available at ScienceDirect

Brain & Language

journal homepage: www.elsevier.com/locate/b&l

The Dutch Linguistic Intraoperative Protocol: A valid linguistic approach to awake brain surgery

E. De Witte [a], D. Satoer [b,c], E. Robert [d], H. Colle [d], S. Verheyen [e], E. Visch-Brink [b], P. Mariën [a,f,*]

[a] Clinical and Experimental Neurolinguistics, Centre for Linguistics, Vrije Universiteit Brussel, Brussels, Belgium
[b] Department of Neurosurgery, Erasmus University Medical Centre, Rotterdam, The Netherlands
[c] Department of Neurolinguistics, Groningen Center of Expertise for Language and Communication Disorders (GELC), University of Groningen, The Netherlands
[d] Department of Neurosurgery, AZ Sint-Lucas, Ghent, Belgium
[e] Department of Psychology, Catholic University Leuven, Belgium
[f] Department of Neurology and Memory Clinic, ZNA Middelheim, Antwerp, Belgium

ARTICLE INFO

Article history:
Accepted 25 October 2014
Available online 17 December 2014

Keywords:
Awake surgery
Intraoperative language testing
Language mapping
Brain tumour
Direct electrical stimulation
Corticosubcortical mapping
Standardised test battery

ABSTRACT

Intraoperative direct electrical stimulation (DES) is increasingly used in patients operated on for tumours in eloquent areas. Although a positive impact of DES on postoperative linguistic outcome is generally advocated, information about the neurolinguistic methods applied in awake surgery is scarce. We developed for the first time a standardised Dutch linguistic test battery (measuring phonology, semantics, syntax) to reliably identify the critical language zones in detail. A normative study was carried out in a control group of 250 native Dutch-speaking healthy adults.

In addition, the clinical application of the Dutch Linguistic Intraoperative Protocol (DuLIP) was demonstrated by means of anatomo-functional models and five case studies. A set of DuLIP tests was selected for each patient depending on the tumour location and degree of linguistic impairment. DuLIP is a valid test battery for pre-, intraoperative and postoperative language testing and facilitates intraoperative mapping of eloquent language regions that are variably located.

1. Introduction

Intraoperative language mapping during awake surgery is considered to be the 'gold standard' since a positive impact of direct electrical stimulation (DES) on postoperative linguistic outcome is generally advocated (De Witt Hamer, Robles, Zwinderman, Duffau, & Berger, 2012; Duffau, Moritz-Gasser, & Gatignol, 2009; Ilmberger et al., 2008; Pereira et al., 2009; Sanai & Berger, 2010; Sanai, Mirzadeh, & Berger, 2008; Spena et al., 2010). DES enables to identify critical cortical and subcortical language areas and pathways, which cannot be resected without post-operative language deficits (De Witt Hamer et al., 2012; Duffau, 2007; Duffau, Lopes, et al., 2005; Spena et al., 2010). During DES, picture naming (Metz-Lutz et al., 1991) and counting are commonly used as language paradigms (Bertani et al., 2009; Duffau, 2007). However, this approach, in which word-finding abilities and automatic speech are investigated in a restricted way, does not cover the integral variety of expressive and receptive language functions, necessary for an adequate communication. Verb–noun association tasks, auditory and visual comprehension tasks, repetition, reading, and writing tasks are occasionally used in experimental settings and some studies described a more tailored selection taking into account tumour location (Bello et al., 2007, 2008; Bertani et al., 2009; Duffau et al., 2003; Duffau, Lopes, et al., 2005) (for a review see De Witte & Mariën, 2013). As a result, test batteries including specific and sensitive tasks are needed for intraoperative language testing in order to preserve language function. Polczynska (2009) suggested three sets of 'home made' tasks consisting of: (1) 'grammar-focused tests', (2) 'non-dominant right-hemisphere tests', and (3) 'tests for subcortical stimulation'. Coello et al. (2013) proposed a minimal common protocol consisting of language tasks such as picture naming, semantic association and reading. In the study of Miceli, Capasso, Monti, Santini, and Talacchi (2012), the following language tasks were included to assess lexical-semantic processing: naming (both objects and actions), lexical decision (both auditory and visual) and word–picture matching (nouns and verbs, auditory and visual). Papagno et al. (2012) suggested a neuropsychological battery (Milano-Bicocca Battery MIBIB) to assess cognition (language, memory, executive functions, apraxia, spatial cognition) in low-grade glioma's pre- and

* Corresponding author at: Department of Clinical & Experimental Neurolinguistics, Vrije Universiteit Brussel, Pleinlaan 2, B-1050 Brussels, Belgium.
E-mail address: peter.marien5@telenet.be (P. Mariën).

http://dx.doi.org/10.1016/j.bandl.2014.10.011
0093-934X/

postoperatively. Intraoperatively, they use naming of objects, actions and people. However, in general little is known about the feasibility, the construction, the validity of the tasks, and no normative data are available. The selection of the type of language paradigms during DES procedures is of crucial importance because of its direct effect on clinical outcome (Hamberger, Seidel, Mckhann, Perrine, & Goodman, 2005). Therefore the sensitivity of each language task used during DES needs to be investigated (Satoer et al., 2012; Talacchi, Santini, Savazzi, & Gerosa, 2011; Talacchi et al., 2012; Teixidor et al., 2007). We attempted to achieve this goal by developing for the first time a Dutch standardised neurolinguistic test battery for awake surgery in critical language areas. The language test battery, called DuLIP (Dutch Linguistic Intraoperative Protocol) includes a variety of phonological, semantic, syntactic and articulatory tests, standardised in a healthy control population of 250 neurologically healthy adults (see part 2 and 3). The clinical use of DuLIP in awake surgery is illustrated with a 'location-function-task' model and this model is applied to select an appropriate intraoperative language test battery to assess linguistic functions in patients with tumoural lesions in eloquent areas (see part 4).

2. Methods

2.1. Procedure

The first two authors of this manuscript and a group of master level students in clinical linguistics, speech and language pathology and clinical psychology were trained to administer the set of linguistic tests of the DuLIP, consisting of one object naming test, four phonological tests (repetition words, phonological odd word out, phonological fluency, phonological sentence judgment), eight semantic tests (semantic odd word out, semantic picture out, semantic association, sentence completions (closed and broad context), semantic fluency animals + jobs, semantic sentence judgment), five syntactic[1] tests (action naming, verb generation, syntactic fluency, syntactic sentence judgment I & II) and one articulatory test (verbal diadochokinesis). 250 healthy volunteers participated in the normative study. All test data were quantitatively (see statistics) and qualitatively analysed.

2.2. Subjects

250 healthy adult volunteers entered the normative study for the DuLIP. Inclusion criteria consisted of (1) Dutch as a mother tongue, (2) no (history of) cardiovascular, neurological, psychiatric, or developmental language and/or speech disorders, (3) no toxic substance abuse (no drug or alcohol abuse), (4) normal vision, (5) normal hearing, (6) no use of sleep medication, psychofarmaca or neuroleptic drugs, and (7) a score above 24/30 on the Mini Mental State Examination (MMSE) (generally accepted cut-off for impairment is 23; Crum, Anthony, Bassett, & Folstein, 1993; Folstein, Folstein, & McHugh, 1975) (http://faculty.pepperdine.edu/shimels/Courses/Files/MMSE.pdf). The participants were informed about the goals of the project and gave a written informed consent. The study was approved by the ethical committee of the Vrije Universiteit Brussel. The number of participants per age group was defined on the basis of current statistics about the prevalence of supratentorial tumours in the Flemish adult population (30% between 18 and 49 years, 55% between 50 and 74 years, 15% 75 years or older (Van Eycken & De Wever, 2006). Both male (18–92 y) and female (18–86 y) participants were included. We recruited subjects from a variety of culturally and philosophically different organizations over different rural and urban regions in Belgium (provinces: West-Flanders, East-Flanders, Antwerp, Flemish Brabant, Limburg). The demographic data of the healthy participants are summarised in Table 1.

Table 1
Demographic characteristics of the 250 healthy participants.

Demographics	Mean (median)	SD	Range
(A) Age, education, MMSE (mean, SD, range)			
Age in years	51.29 (54.00)	19.81	18–92
Education in years	13.78 (14.00)	3.27	6–23
MMSE	29.16 (30.00)	1.18	24–30
Demographics	**Groups**	**Number of subjects**	**Percentage**
(B) Gender, age, education, province, handedness groups			
Gender	M	101	40.40
	F	149	59.60
Age	18–54 y	129	51.60
	55+ y	121	48.40
Education level	⩽12 y	104	41.60
	>12 y	146	58.40
Province	West Flanders	44	17.60
	East Flanders	114	45.60
	Antwerp	20	8.00
	Flemish Brabant	40	16.00
	Limburg	32	12.80
Handedness	L	13	5.20
	A	8	3.20
	R	229	91.60

(A) Legend: SD = standard deviation; MMSE = Mini Mental State Examination.
(B) Legend: M = male, F = female, y = year, L = left-handed, A = ambidexter (Laterality Quotient ⩾−40 en ⩽+40), R = right-handed.

2.3. Test administration, registration and correction

The tests were administered in an identical order. Practice items were included to ensure that participants understood the task. Visual stimuli were presented in a powerpoint presentation on a laptop screen (Table 2). A division was made between tasks for use during DES stimulation within 4 s (A) and tasks for use during resection (no time restriction) (B). Since corticosubcortical stimulation may maximally last 4 s, all language tasks should ideally be performed within this time frame. However, certain tasks measuring important language aspects such as the fluency tasks (language initiation) and judgment tasks (language perception) are not feasible within 4 s. Since these tasks provide valuable information they were included in the protocol and used during resection in between stimulations. Of course, the tasks for stimulation (A) are also very relevant to apply during resection. Test items for use during DES were presented every 4 s accompanied by a beep called 'laser sound'. To avoid fatigue the DuLIP protocol was assessed in two parts (A & B) at two different test moments. The total length of both assessments (heteroanamnesis, EHI, MMSE, DuLIP) was 1.5 h. The answers were recorded verbatim and transcribed. One point was given if the target answer was correctly produced and within the given time frame. Self-corrections were allowed if given within the 4 s time frame. One repetition of an auditory task on request of the participant was allowed. The accuracy of the scoring of the tests was checked by one of the first two authors and was based on qualitative terms agreed on a priori. Uncertainties regarding the correctness of test items were discussed in group with the other authors until a consensus was reached.

2.4. Description of tests

The phonological, semantic, syntactic, naming tasks and articulatory task of the DuLIP are presented in Table 2. Taking into account the constraints of the awake procedure (fixed position, limited time) we carefully selected existing language tests

[1] The term syntax is used to define tasks that measure grammatical aspects.