Universidade de Aveiro Escola Superior de Saúde
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THE SPEECH SOUND SYSTEM IN AWAKE BRAIN SURGERY: A PHONOLOGICAL AND ARTICULATORY EVALUATION

Os Sons da Fala em Cirurgia Cerebral Acordada: Avaliação Fonológica e Articulatória

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Tese apresentada à Universidade de Aveiro para cumprimento dos requisitos necessários à obtenção do grau de Mestre em Terapia da Fala, realizada sob a orientação científica do Doutor Luís Miguel Teixeira de Jesus, Professor Coordenador com Agregação da Universidade de Aveiro.

The brain is wider than the sky,

For, put them side by side,

The one the other will include

With ease, and you beside.

The brain is deeper than the sea,

For, hold them, blue to blue,

The one the other will absorb

As sponges, buckets do.

The brain is just the weight of God,

For, lift them, pound for pound,

And they will differ, if they do,

As syllable from sound.

Emily Dickson

O júri

Presidente	Professora Doutora Marisa Lobo Lousada Professora Adjunta da Universidade de Aveiro
Arguente	Professor Doutor João Manuel Pires da Silva e Almeida Veloso Professor Associado com Agregação da Universidade do Porto
Orientador	Professor Doutor Luís Miguel Teixeira de Jesus Professor Coordenador com Agregação da Universidade de Aveiro

Acknowledgements

I would like to express my deepest appreciation to:

Professor Luís Jesus, for proposing these challenges and pushing me to accomplish more than I thought possible. Thank you for your constant trust and motivation, which helped me to overcome the difficulties of this ambitious project.

The original authors of the DuLIP, Evy Brink, Djaina Satoer and Elke De Witte, for trusting our team with such an important project and for the availability you always showed.

Mafalda and Mariana, who shared this journey with me, with all its difficulties but also victories and successes. Thank you for being my teammates, but most importantly, my friends.

Doctor Catarina Viegas, for the amazing opportunity to watch her work and for believing that speech and language therapy can be a decisive factor in the recovery of patients.

Professor Andreia Hall for the help she gently provided in the statistical analysis.

Isabel Monteiro da Costa and Assunção Matos, who inspire me professionally and personally since the beginning of my academic journey. You are a breath of fresh air in the practice of speech and language therapy.

My work colleagues and friends for their attention and encouragement. Thank you for always listening to my worries and excitements.

Fábio, as we grow together. Thank you for all the support and encouragement, and especially for the English translation of this work. I will repay in kwanzas.

My parents, for allowing me to have what they never could, and to my brother. You are the best examples of resilience I have. Thank you for being so supportive and for giving me the stability required to keep up with my work.

Everyone who participated in this study, for their patience and availability.

Palavras-chave Cirurgia Cerebral Acordada; Gliomas de Baixo Grau; Fonologia; Articulação; Linguagem; Fala; Avaliação.

Resumo

Uma avaliação adequada da fala e linguagem em cirurgia cerebral acordada tem implicações funcionais que se repercutem na qualidade de vida do paciente oncológico. Em Portugal, não são conhecidos instrumentos padronizados e validados para a avaliação destas competências em cirurgia cerebral acordada. Para colmatar essa necessidade, este estudo visa adaptar ao Português Europeu as provas dos domínios fonológico e articulatório do Dutch Linguistic Intraoperative Protocol (DuLIP) e contribuir para a sua padronização. Em todo o processo foi fundamental o apoio de uma equipa multidisciplinar. Após a adaptação dos estímulos, um estudo normativo foi realizado num grupo de 144 indivíduos saudáveis. Foram estudados os efeitos da idade, género e nível de escolaridade nas tarefas adaptadas. Foram efetuados cálculos para medir a correlação entre as capacidades de atenção e memorização e a prova de identificação do intruso fonológico. Apenas as tarefas que apresentaram uma percentagem de acerto superior a 90% foram mantidas na versão final do instrumento. O nível de escolaridade exerceu influência sobre a prova de fluência fonológica ($p<.001$) e o género sobre a prova de repetição ($p=.002$). Este é um estudo inovador, pois constitui parte integrante da primeira contribuição para a padronização de um instrumento a ser utilizado em cirurgia cerebral acordada em Portugal. Salvaguarda-se a necessidade de cumprir outras etapas para a sua utilização clínica.

Keywords Awake Brain Surgery; Low-Grade Gliomas; Phonology; Articulation; Language; Speech; Evaluation;

Abstract

An adequate evaluation of language and speech during an awake brain surgery has functional implications that can affect the quality of life of oncologic patients. In Portugal, there is a lack of standardised and validated tools to conduct this type of evaluation. To address this need, this study seeks to adapt the phonological and articulatory tasks of the Dutch Linguistic Intraoperative Protocol (DuLIP) into European Portuguese, while also contributing toward its standardisation. To achieve this, the support of a multidisciplinary team was required. After adapting the stimuli, a normative study was conducted in a group of 144 healthy individuals. The impact that the age, gender and years of schooling of these individuals had on the adapted tasks was measured. Calculations were made to examine if attention and memorisation levels correlate with the phonological odd word out task. Only tasks with a hit rate higher than 90% were included in the final version of the tool. The years of schooling had a significant influence in the phonological fluency task ($p<.001$) and the gender also had a significant impact in the word repetition task ($p=.002$). This is an innovative study in Portugal because it is one of the first contributions to the standardisation of a tool that can be used during an awake brain surgery. However, additional steps are required before this tool can be used in the clinical practice.

Abbreviations and acronyms

ACE-III – Addenbrooke's Cognitive Examination III

AF – Arcuate Fascicle

AMR – Alternating Motion Rate

CVA – Cardiovascular Accident

DES – Direct Electrical Stimulation

DuLIP – Dutch Intraoperative Linguistic Intraoperative Protocol

DuLIP-EP – Dutch Intraoperative Linguistic Intraoperative Protocol – European Portuguese

EP – European Portuguese

MMSE – Mini-Mental State Examination

SLF – Superior Longitudinal Fascicle

SMG – Supramarginal Gyrus

SMR – Sequential Motion Rate

vPMC – Ventral Premotor Cortex

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1. Introduction

Awake brain surgery has been increasingly used in the last few years. First mentioned at the end of the 19th century, this surgery went through important refinements when Ojemann (1979) showed that the areas associated with the linguistic process aren't located solely in the perisylvian language zone, but in a broader neuronal network, varying from one person to another (Bizzi, 2009; Borchers, Himmelbach, Logothetis, & Karnath, 2012; July, Manninen, Lai, Yao, & Bernstein, 2009; McDermott, Watson, & Ojemann, 2005). This surgery is primarily performed in patients with epilepsy or brain tumours in eloquent areas, i.e., areas that once removed, result in postoperative deficits (Ruis, 2018). With regard to the tumours located in eloquent areas of speech and language, awake brain surgery, alongside neurophysiological evaluation and intraoperative mapping, is one of the preferential lines of treatment (Darder & Lopez, 2012). Current guidelines suggest that the surgery should aim for the greater extent of resection to maximise postoperative gains (Forst, Nahed, Loeffler, & Batchelor, 2014; Pereira et al., 2009; Sepúlveda-Sánchez et al., 2018).

Patients who undergo this surgery must meet a set of inclusion and exclusion criteria that although not formally and universally defined, gather a general consensus in the literature. These criteria must encompass the characteristics of the tumour (histology and location) and the personal characteristics of the patient (Coello et al., 2013). Regarding the former, the use of awake brain surgery is most frequent in gliomas, which are the most common type of primary brain tumour, emerging from the glial cells in the central nervous system. These can be classified as astrocytomas, oligodendrogliomas, mixed oligoastrocytomas and ependymomas. As for the latter, it is necessary to consider the prior cognitive level, age, psychological characteristics and levels of activity and participation of the patient. Old age, cognitive impairments, overweightness and psychiatric and psychological disorders are considered exclusion criteria for this surgery (Bertani et al., 2009; Sepúlveda-Sánchez et al., 2018; Van Den Bent, Snijders, & Bromberg, 2012).

Direct Electrical Stimulation (DES) is considered the gold-standard surgical procedure during awake brain surgery. It allows the mapping of the brain during the surgery, which in turn enables the identification of cortical and subcortical areas and pathways that are fundamental for language. It is a method that assists in risk/benefit decision making, and it has a positive impact on the survival time and quality of life of the patients, as it enables a greater remission of the tumour area with fewer deficits associated with eloquent areas. It is therefore referenced as the best method to maximise the 'onco-functional balance' (Coello et al., 2013; Rofes et al., 2017), as it seeks to prevent the patient from losing a certain level of functionality (Bizzi, 2009; Coello et al., 2013; De Witt Hamer, Robles, Zwinderman, Duffau, & Berger, 2012; Duffau, Moritz-Gasser, & Gatignol, 2009; Pereira et al., 2009; Sanai, Mirzadeh, & Berger, 2008).

Despite this procedure allowing a more precise identification of the language pathways, Functional Magnetic Resonance Imaging and Diffusion Tensor Imaging are important imaging techniques to aid the surgical approach, and can even reduce the duration of the surgery and contribute to better outline the remission area (Bello et al., 2010; Duffau, 2005; Mandonnet,

Winkler, & Duffau, 2010; Young, Brennan, Fraser, & Brennan, 2010). These techniques should take place during various moments: Pre-surgery, intra-surgery and post-surgery (Bertani et al., 2009).

The patient has an active role in this surgery, therefore a series of specificities regarding the anaesthesia administered are taken into consideration (Witte, 2015). Pereira et al. (2009) mention the use of local anaesthesia, which allows the application of an asleep-awake-asleep method. The first phase of this method is the correct positioning of the patient by the team. Next, the patient's head is secured using a Mayfield clamp and a craniotomy is performed (Pereira et al., 2009). Before opening the dura mater, the patient is woken and the intraoperative brain mapping is conducted. Direct Electrical Stimulation is performed with a bipolar stimulator that produces biphasic pulses with a duration of 1 ms each and a frequency of 25-60 Hz. In order to evaluate linguistic disturbance, the stimulation should be no longer than 4 s, preventing epileptic seizures. The cortico-subcortical areas are identified and after removing the tumour, the patient is reintubated and the dura mater, cranium and scalp are closed (Duffau, 2007, 2014; Szelényi et al., 2010).

A patient with a tumour in speech and language areas who has been selected to undergo this surgery must be submitted to an intraoperative assessment of the linguistic skills to be tested (Witte, 2015), in addition to the DES, which allows temporary disabling certain structures at the cortical and subcortical level (Duffau, Gatignol, Mandonnet, Capelle, & Taillandier, 2008). Language is traditionally defined as the result of a 'complex nervous activity that enables intraindividual communication of psychic states through the materialisation of multimodal signs symbolising these states, according to the conventions of a linguistic community' (Lecours, Lhermitte, & Bryans, 1983). It is a skill that permits the storing, evoking and combining of symbols in an 'endless exchange of expressions that allows the elaboration of thought'. It encompasses various domains: Semantic, morphosyntactic, phonological and pragmatic (Antonsson, 2017; Resende, Reis, & Magalhães, 2003).

The evaluation of speech and language is one of the main responsibilities of a Speech and Language Therapist (APTF, 2019). When this evaluation is performed in a surgical context, they play an important role in the neurosurgery team, assessing the preservation of the domains associated with these eloquent areas. This role includes implementing and interpreting the results of speech and language tasks in the pre-, intra- and postoperative moments, following along the intervention process (Geemen, Herbet, Moritz-gasser, & Duffau, 2014). In the intraoperative period, speech and language therapists are an important asset because they report in real-time the alterations in the patient's speech, such as any associated degree of dysarthria, speech arrest and alterations in facial movements and language, like perseverations or paraphasias, while also providing their respective classifications (Geemen et al., 2014; Maldonado, Moritz-Gasser, & Duffau, 2011). Furthermore, having a Speech and Language Therapist in the neurosurgery team is beneficial because their interaction with the neurosurgeon and the rest of the team is determinant for the functional optimisation of the results of the process (Rofes et al., 2017).

In Portugal, the language evaluation of adults is mainly conducted with the use of informal tools created by the speech and language therapists for personal use, as well as the Lisbon Aphasia

Examination Battery (Leal, 2009). However, the tests in use, developed in Portugal and even internationally, are fairly limited in their evaluation of language skills during the intraoperative cortical mapping (Połczyńska, 2009). In this surgery, several technical-contextual specificities are considered, and the evaluation must be performed accordingly. The time required to present and respond to the stimuli (4 s - duration of the DES), the fact that the head is in the same fixed position for hours, the space limitations for the equipment, and the patient's stress and discomfort, are important restrictions to consider. Moreover, low-grade gliomas are characterised as progressing slowly, although they can evolve into high-grade gliomas, which are more aggressive and grow faster. Nevertheless, this fact highlights the need to use a fairly sensitive test to evaluate the preservation of language skills in these cases, since neuroplasticity can lead to some unnoticed deficits. For that very same reason, it is common for these patients to show less severe deficits than those who acquire aphasia, e.g., after a cerebrovascular accident (CVA) (Antonsson, 2017; Desmurget, Bonnetblanc, & Duffau, 2007; Geemen et al., 2014). This shows the importance of having a tool designed for this specific context, which is so distinct because of all the reasons mentioned above (Kanno & Mikuni, 2015).

The Dutch Linguistic Intraoperative Protocol (DuLIP) is a Dutch standardised neurolinguistic battery structured to comply with the specificities of the awake brain surgery. It includes a variety of tasks from the areas of phonology, semantics, syntax and articulation and it was standardised in 250 healthy individuals. Its clinical use is described as a 'location-function-task' model that is applied to evaluate linguistic tasks in an intraoperative setting. Nonetheless, this battery of evaluation encompasses the gathering of data during three moments: Pre-, intra- and post-surgery. Moreover, it covers the various domains of comprehension and expression of language and it is designed to be applied in the operating room and to detect small variations that might go unnoticed in a regular language evaluation test (Miceli, Capasso, Monti, Santini, & Talacchi, 2012; Witte et al., 2015).

This is an extensive battery and therefore the tasks to apply must be selected prior to the surgery based on the characteristics of the tumour and the patient (Witte et al., 2015). This Thesis in particular focuses on the phonological and articulatory areas of the DuLIP.

Phonology is the area of language sciences responsible for studying the sound. It focuses on studying sound abstractly and the corresponding adjacent processes, i.e. it studies the system of rules of the language which determines the organisation of sounds in syllables and words (Stemmer & Whitaker, 2008). Articulation is one of the components of speech and represents the neuromuscular ability required to achieve verbal output (Brookshire, 2015; Gill & Damann, 2015). A disturbance in each of the domains mentioned above can lead to a different diagnosis. Thus, a disturbance in phonological processing is considered a disturbance in language and is frequently a symptom of aphasia (Stemmer & Whitaker, 2008), traditionally identified by a disturbance in linguistic ability provoked by brain damage (Ardila, 2006). As for the difficulty in motor planning and execution for speech, it is considered a phonetic problem, i.e., articulatory. A change in motor execution for speech results in dysarthria. Dysarthria is a motor speech disorder in which the speech subsystems (respiration, phonation, resonance, and articulation) are affected (Stemmer & Whitaker, 2008). Another possible cause for articulatory alterations is speech apraxia, in those cases where changes arise from the difficulty in the motor programming for speech (Resende et al., 2003). The etiology of these disturbances can point to an acute

episode or a progressive condition, caused, e.g., by CVA, traumatic brain injury, neurological diseases or brain tumours (Ardila, 2006).

Despite inter-individual variability, each of the domains of speech and language is associated with specific brain areas (Friederici, 2011). In this regard, many studies correlate the triggering of brain areas with domains of language and tasks carried out during surgery.

More specifically, regarding the phonological domain, the cortical influence becomes evident at the level of the Supramarginal Gyrus (SMG), located in the parietal lobe, Brodmann area 40, because when this is stimulated, phonological alterations occur in naming and repetition tasks (Duffau, 2007; Duffau et al., 2005; Maldonado et al., 2011; Moritz-Gasser & Duffau, 2013). The work of Maldonado et al. (2011), specifies the triggering of this structure in tasks of phonological judgment (Maldonado et al., 2011). Moreover, the stimulation of the inferior part of the SMG induced anomia and phonological disorders (Moritz-Gasser & Duffau, 2013).

The temporal lobe is also involved in tasks from this domain (Duffau, 2007), with the middle portions of the temporal lobe being involved in processing speech signals for comprehension (Hickok, 2012). The stimulation of the posterior (the so-called Wernicke's area), superior and middle temporal gyri induced anomia and perseverations in the word repetition task (Coello et al., 2013).

The Superior Longitudinal Fascicle (SLF) and its respective cortical projections (inferior frontal cortex, inferior parietal lobe and temporal lobe) are also involved in this domain, since when stimulated, they cause phonemic paraphasias (Coello et al., 2013). The Arcuate Fascicle (AF) is a structure commonly involved in phonological tasks (Duffau, 2007). Many studies show how important the subcortical structure is for this domain, since when stimulated, it is common to find phonological disorders such as phonemic paraphasias in word repetition tasks, especially in the posterosuperior portion of the AF (Coello et al., 2013; Maldonado et al., 2011; Moritz-Gasser & Duffau, 2013).

In phonological fluency tasks, the frontal cortex, e.g., in Brodmann areas 4, 6 and 44, is vital to phonemic evoking (Duffau et al., 2005). Furthermore, during this activity, the anterior temporal regions, left inferior parietal cortex (frequently associated with working memory), the insula and putamen are triggered (Baldo, Schwartz, Wilkins, & Dronkers, 2006; Duffau, 2007).

Regarding articulation, this ability involves the ventral premotor cortex (vPMC), i.e. the anterior segment of the lateral part of the SLF (Maldonado et al., 2011). Its inactivity provokes anarthria, i.e. the loss of motor ability to speak, (Geemen et al., 2014), and dysarthria, revealing the presence of speech arrest (Duffau et al., 2005; Moritz-Gasser & Duffau, 2013). The vPMC is an area with limited plasticity and therefore of important preservation (Geemen et al., 2014; Maldonado et al., 2011). Moreover, the anterior part of the SLF leads to articulatory alterations (Coello et al., 2013; Moritz-Gasser & Duffau, 2013). The same study demonstrates the influence of the dominant supplementary motor area, anterior insula, lentiform nucleus and vPMC in articulation; it was possible to diagnose a dysarthria when these areas were stimulated. Duffau (2007), indicates that the role of the lentiform nucleus is associated with motor planning for speech (Duffau, 2007). Additionally, disturbances in motor programming have been linked to the subcallosal fasciculus (Coello et al., 2013).

Bello et al. (2007) mention the role of Broca's area in this domain because when stimulated, it induces speech arrest. They further mention that the stimulation of the anterior portion of the body of the lateral ventricle induced dysarthria and that the stimulation of the area corresponding to the head of the caudate induced perseverations. They also indicate that the stimulation of the white matter close to the anterior border of the insula medial to Broca's area induced phonemic paraphasias, and higher areas may result in speech arrest (Bello et al., 2007). Regarding the role of the insular region in articulatory tasks, the anterior insular region is associated with the articulatory coordination ability (Baldo et al., 2006) and the descending pathway, which connects the anterior insula and the vPMC to the primary sensorimotor area of the mouth and eventually joins to the pyramidal tract, also plays a major role. The stimulation of these fibres causes anarthria (Bello et al., 2010; Coello et al., 2013; Duffau, Taillandier, Gatignol, & Capelle, 2006).

1.1 Aims of the study

This study aims to: 1) to culturally adapt the DuLIP's phonology tasks; 2) culturally adapt the DuLIP's articulatory task; 3) standardise DuLIP's phonological tasks to the European Portuguese (EP) normal population; 4) standardise DuLIP's articulatory task to the EP normal population; 5) study if the components of attention and memorisation are correlated with the phonological odd word out task of the DuLIP-EP (Dutch Linguistic Intraoperative Protocol - European Portuguese), since language is not an ability that exists in isolation from other brain functions, but is related to and dependent on other functions including hearing, vision, attention, memory and motor ability (Resende et al., 2003).

2. Method

The study conducted comprises two parts: The linguistic-cultural adaptation of the DuLIP and a contribution towards the standardisation of this test battery for the Portuguese population. To accomplish these goals, data were quantitatively and qualitatively analysed through descriptive statistics and study of correlations (Fortin, 1999).

All ethical guidelines were followed. This study was approved by the Ethics Committee of the Research Unit of Health Sciences at the School of Nursing in Coimbra (UISCISA reference: 535/01-2019, Coimbra, Portugal (Appendix IV).

2.1. Adapting the DuLIP to European Portuguese

The translation and adaptation of language assessment tools include some challenges: The cultural differences between languages; differences in the access to databases with information on linguistic attributes (i.e. word frequency and imageability); and differences specific to each domain of the language, e.g., spelling-to-sound regularity with regard to phonology (Fyndanis et al., 2017).

In order to solve these challenges, more than a literal translation was necessary to culturally and linguistically adapt the DuLIP according to the linguistic variables that were in its origin. The original authors provided the stimuli used in Dutch and their English translation. Following their initial suggestion, the adaptation process began with the literal translation of the stimuli in English, freely and individually done by three members of a workgroup. The material resulting from this phase was discussed within the group, in an expert meeting, to gather consensus for a pilot version. Because a series of items failed to respect the psycholinguistic characteristics at the basis of creation of the original stimuli, the resulting materials were then individually adjusted and adapted according to those same properties. Additionally, there was an attempt to avoid stimuli with emotional connotation and to keep the original items, whenever possible.

It should be noted that, alongside this work, there are other groups involved, from Chile, France, Singapore and Austria. The same adaptation protocol was used for the different languages. The EP team was composed of the original authors of the DuLIP, one neurosurgeon, three speech and language therapists, one of them the author of this Thesis, and the Thesis' supervisor. The other two speech and language therapists included were those who adapted the semantic (Cardoso, 2019) and syntactic (Martins, 2019) domains of the DuLIP.

Regarding the phonological component, there are tasks of word repetition, phonological odd word out, phonological sentence judgment, and phonological fluency. As for articulation, there is a task of verbal diadochokinesis. Each of these tasks has its own specific properties, which are mentioned below.

Word repetition: This task allows the evaluation of speech word production through the phonological input and output route (Witte et al., 2015). This skill plays a crucial role in language

development by enabling the learning of new words. Clinically, word repetition is included in the classification of aphasic diagnosis and can provide important prognosis information (Hosomi et al., 2009) and guide rehabilitation (Moritz-Gasser & Duffau, 2013; Schlaug, Marchina, & Norton, 2009). Additionally, it involves several levels of linguistic processing. The conversion of phonological information into verbal articulation requires various steps: 1) phonological decoding and temporary storing inside the working memory system; 2) semantic recognition of the word; 3) phonological coding; and 4) motor execution and planning (articulatory component) (Moritz-Gasser & Duffau, 2013).

For the adaptation of this task, the principal criteria were the word complexity levels, with 6 complexity levels taken into consideration to increase the difficulty at the phonological and articulatory levels: 1) 3-syllable words without consonant clusters and without phonemic similarities; 2) 3-syllable words with consonant clusters and without phonemic similarities; 3) 3-syllable words without consonant clusters and with phonemic similarities; 4) 3-syllable words with consonant clusters and with phonemic similarities; 5) 2-syllable words without consonant clusters and without phonemic similarities; 6) 2-syllable words with consonant clusters and without phonemic similarities (Witte et al., 2015).

The number of items per complexity level should match the original count, with 75 words for complexity level 1; 50 words for complexity level 2; 40 words for complexity level 3; 10 words for complexity level 4; 100 words for complexity level 5; and 25 words for complexity level 6. The total number of items for this task is 300 words. These levels did, therefore, vary regarding the presence of consonant clusters and phonemic similarities. Many studies were at the basis of this division by showing that words with consonant clusters and similar phonemes (especially vowels) are more prone to errors (Béland, Paradis, & Bois, 1993; Galluzzi, Bureca, Guariglia, & Romani, 2015; Maas, Barlow, Robin, & Shapiro, 2002; Nespoulous & Moreau, 1998). The phonemic similarities can also refer to syllables that are non-consecutive, and therefore words like [li.miˈar] and [pruˈtɛʃ.tu] were included.

In the adaptation process, words whose literal translation fit in the original complexity level were kept in that same level (e.g. the Dutch word <gorilla>, English translation <gorilla> and Portuguese translation <gorila>, was able to be kept in the same complexity level, because there are no changes in the number of syllables, presence of phonemic similarities and presence of consonant clusters). If the literal translation of a word matched another complexity level, the word was changed to that same level (e.g. the Dutch word <tijger>, whose English translation is <tiger> and Portuguese translation is <tigre>, the word was moved into a different complexity level, because although it maintains the same number of syllables, the Portuguese translation encompasses a consonant cluster that does not exist in Dutch, i.e. the complexity level increases). It is also possible that a certain word does not fit into any complexity level, and is thus removed and replaced by another that is present in the Portuguese language and that meets the above-mentioned psycholinguistic properties. Table 1 presents some examples of this task and how it is organised in terms of complexity.

Other than the complexity levels, some words were also considered because of their diversity in terms of frequency (data extracted from the Reference Corpus of Contemporary Portuguese [RCCP]), number of syllables and number of phonemes. Concerning the latter, all of the

phonemes of EP were considered, in the possible positions of the word. In this regard, the sound inventory of European Portuguese has nine oral vowels, five nasal vowels, two glides, six stops, three nasal consonants, one trill, one tap, eight fricatives and two laterals. With the exception of /ɲ/, /ɾ/ and /ʎ/, all other phonemes can occur at the start of a word. Phonologically, only /l/, /ɾ/ and /ʃ/ can occur at the end of a word (Jesus, Valente, & Hall, 2015; Mateus, Falé, & Freitas, 2005).

Table 1: Word repetition task (examples)

Portuguese word	English translation	Number of syllables	Phonemic similarities	Consonant cluster	Complexity level
<lagarto>	*<lizard>*	3	*X*	*x*	1
<tradição>	*<tradition>*	3	*X*	[**tr**ɐ.diˈsɐ̃w̃]	2
<ananás>	*<pineapple>*	3	[**ɐ**.n**ɐ**ˈnaʃ]	x	3
<lavrador>	*<farmer>*	2	[l**ɐ**.vr**ɐ**ˈdor]	[lɐ.**vr**ɐˈdor]	4
<mesa>	*<table>*	2	*X*	*x*	5
<blusa>	*<'blouse>*	2	*X*	[ˈ**bl**u.zɐ]	6

Phonological odd word out: The goal with this task is to evaluate the component of metalanguage at the phonological level via the lexical input route. This task was eliminated from the original tool since the authors determined that it should be refined in order to better assess the function targeted. The justification for mentioning this limitation in the original study was based on the fact that the stimuli were presented visually and therefore the phonological odd word out task could be too easy since 'some answers can be visually identified without using phonological knowledge (e.g. bal, hal, val, weg)' (Witte et al., 2015). However, for the EP version, it was decided that the stimuli would only be presented auditorily and that this task would be kept in the DuLIP-EP.

To present words with a equal number of phonemes, the syllabic structure of each of the items was controlled (e.g. [ˈmɛl, ˈfɛl, ˈʒɛl,ˈmil] – all words are monosyllabic and maintain a CVC syllabic structure). Accordingly, 15 items were included in this task. Each of them contains 4 monosyllabic words that rhyme with each other.

Phonological sentence judgment: This task aims to assess phonological awareness by asking the participants to point out which of the auditorily presented sentences is phonologically correct and incorrect. Additionally, this task measures phonological decoding and verbal short-term memory (Witte et al., 2015).

As in the original test, a total of 30 items was used in this task. Initially, the sentences were translated literally. If the pseudowords used did not respect the rules of the Portuguese language, e.g. the syllabic structure and the position of the phonemes in the syllable, they should be substituted for others that did. Because it was not possible to find a database of pseudowords for EP, the stimuli were created from scratch. For this task, the characteristics of the sentence, such as the mode (declarative), verb tense and voice (passive or active) used, were taken into

account. The phonological similarities and alliterations present in the sentence should also be registered, like in the original study. It should also be mentioned that in the adaptation to EP, the pseudowords must take the same place in the sentence that they took in the original stimulus (e.g. if the pseudoword is in the place of a name in the original stimulus, it takes the place of a name; if it is in the place of a verb, it takes the place of a verb). Sentences whose goal was to be considered correct (n=15) were translated literally from the English version. For example, the sentence <The chocolates tasted delicious>, whose Portuguese translation is <Os chocolates estavam deliciosos>, was meant to be considered correct, so no changes were made. On the other hand, the sentence <The schokel does a dulletje>, which was meant to be considered incorrect, has inaccuracies in the formation of the pseudowords because these do not meet the phonological principles of EP, e.g., in the phonemic position. Therefore, the sentence above was adapted into <O cromel faz um jagole>, keeping the active voice, declarative mode, present simple tense, number of words and SVO structure.

Phonological fluency: The aim of this task, which requires generating words with a given phoneme, is to assess the flexibility of phonological thought and internal driven language. The phonemes /p/, /m/ and /ʀ/ were chosen for the EP version (Cavaco et al., 2013). This task evaluates the ability to evoke phonemes and requires several cognitive domains such as lexical and phonological memory, self-monitoring and cognitive flexibility (Baldo et al., 2006; Satoer et al., 2013).

Verbal diadochokinesis: A verbal diadochokinesis task was used to evaluate the articulatory domain. This task aims to evaluate articulatory performance, i.e. the planning, coordination and execution abilities of the motor speech system by measuring the ability to repeat a segment of speech at high rate It is one of the recommended tasks to evaluate neurological disorders (Devadiga & Bhat, 2012). Alternating motion rate (AMR) and sequential motion rate (SMR) are the two traditional tests of oral diadochokinesis used to assess motor speech production. AMR involves a single syllable being repeated as fast as possible, whereas for SMR a sequence of syllables is repeated as fast as possible. The syllables traditionally employed are [pa], [ta] and [ka] for AMR and the sequence [pataka] for SMR (Pierce, Cotton, & Perry, 2013).

There was no need to adapt this task to EP because the original stimuli contain not only the production of anterior sounds, which involve the orbicular musculature of the lips, but also the tongue tip and back tongue sounds, in which other muscles are involved (Pierce et al., 2013). Moreover, the DuLIP includes the most common sounds for the evaluation of this task (Devadiga & Bhat, 2012).

Some of the adapted tasks can be found in Figure 1.

<Mel> (<honey>)	**<O gaco corre para a Lirdo>**	**[papapa]**
<Fel> (<gall>)	<A tempestade estragou as vindimas>	**[pataka]**
<Gel> (<gel>)	**<O cromel faz um jagole>**	**[paftafkaf]**
<Mil> (<one thousand>)	<A roupa está pendurada na corda>	
A	**B**	**C**

Figure 1: (A) Phonological odd word out. The target item is <*mil*> (one thousand) because it is the only word that does not rhyme with the others; **(B) Phonological sentence judgment.** The target is to validate the sentences: the first and the third sentences as wrong and the second and fourth sentences as correct; **(C) Verbal diadochokinesis.** The goal is to produce the sequences as fast as possible.

2.2. Applying the DuLIP-EP to the general population

2.2.1. Participants

The DuLIP-EP normative study was applied to 144 healthy adult volunteers. The participants had to be over 18 years old, with no upper age limit. The following inclusion criteria were defined: 1) European Portuguese as a mother tongue; 2) no record of cardiovascular, neurological, psychiatric or speech and language disorders; 3) no record of substance abuse; 4) normal vision; 5) normal hearing; 6) no use of sleep induction, psychopharmaceutical or neuroleptic drugs; 7) a score above 24/30 on the Mini-Mental State Examination (MMSE). For this test, the general impairment threshold is 23 points (Folstein, Folstein, & McHugh, 1975). Because the MMSE has not been validated for use in EP, Addenbrooke's Cognitive Examination III (ACE-III) was used instead (Peixoto, Beata, & Pimentel, 2013). The scores were then converted to match the MMSE (Matías-Guiu et al., 2018) with the goal of using the same criteria employed in the original study.

Before partaking in the study, the participants were informed about the objectives and gave written informed consent (Appendix I). The selection of the participants was based on their availability to take on the test (convenience sample).

2.2.2. Data collection

Gathering of data took place between April and August of 2019. Each test had a duration of around one hour and a half per person and included 1) anamnesis (Appendix II); 2) Addenbrooke's Cognitive Examination III (ACE-III); and 3) the DuLIP-EP.

Data were collected by three speech and language therapists who followed specific and previously defined instructions. Example items were included in all tasks to ensure that all participants understood the proposed tasks.

2.2.3. Procedures and assessment tools

The questions were answered orally and the answers were written down by the speech language therapists. The anamnesis included the participants' personal, sociodemographic and clinical data, which helped meet the inclusion criteria on one hand and eased the characterisation of the sample on the other. The ACE-III was applied to rule out any cognitive impairments. The application and grading of this test followed all the corresponding instructions (NeuRA, 2012). Finally, the DuLIP-EP was applied. If the participants showed any signs of fatigue, the test was interrupted and postponed to a later date, if necessary. To grade the tasks in the DuLIP-EP, a point was given for each correct answer that respected the time limit. The assessor could repeat the query once, if necessary. When the assessors had any doubts about how to grade a test, the working group discussed these questions until a consensus was reached. Self-corrections were allowed and considered correct as long as they respected the time limit (4 s).

In the DuLIP, the tasks are split into two groups: time-limited and not time-limited. Because the cortico-subcortical stimulation must not exceed 4 seconds, the language tasks should all be performed within this time frame. However, some components of language such as initiation and perception cannot be evaluated within 4 seconds (e.g. in fluency and judgment tasks, respectively). Nonetheless, since these tasks also provide valuable information, they were included in the protocol and used during the resection of the tumour, between the stimuli. The time-limited stimuli presented via PowerPoint are complemented by a beep called 'laser sound', which helps to identify when the stimulus changed (Witte et al., 2015). All tasks are included in Table 2, where they are grouped according to their time limit (4 s) or lack thereof.

Table 2: Phonological and articulatory tasks of the DuLIP

Timing of assessment(s)	Linguistic level	Task
During DES (in 4 s)	Phonology	- Word repetition • 3-syllable words without CC and PS • 3-syllable words with CC and without PS • 2-syllable words without CC and PS • 2-syllable words with CC and without PS • 3-syllable words without CC and with PS • 3-syllable words with CC and PS
	Articulation	- Verbal diadochokinesis*
Not during DES (without time frame)	Phonology	- Phonological sentence judgment - Phonological odd word out - Phonological fluency

*presented with PowerPoint slides; CC = Consonant cluster; PS = Phonemic similarities

The phonological tasks were not administered consecutively, instead being intercalated with tests from the other domains of language. However, the first task to be presented from the phonological domain was the word repetition task. In this task, the person providing the stimuli read the word with good diction and projection while stressing the tonic syllable. Lip reading was avoided. Participants were asked to immediately repeat 2 and 3-syllable words. Phonologically or semantically related words were not consecutively presented to avoid perseveration.

Afterwards, the phonological odd word out task was presented. This task is presented orally, i.e. without any physical materials, to avoid giving visual cues (i.e. the ending of the words with the same set of graphemes can give away its relation with the rime). Accordingly, the participants listened to a list of four words where one of the words did not rime with the others. The assessor must read all the words with the same prosody, without emphasising the intruder. This required some training before administering the task. The participant was then asked to identify which of the words did not rime with the others.

Six tasks from the other domains (semantic and syntactic) were subsequently carried out and the verbal diadochokinesis task was applied. For this task, the participants were asked to audibly enunciate nine monosyllable sequences as fast and precisely as possible. To help with the automatic monitoring of this task, there was an associated PowerPoint presentation where the sequences to enunciate were saved. Two practice items were used for demonstration and training purposes. The assessor could help to count the sequences enunciated until the fifth one. Time was measured and recorded.

There was then a naming test and a phonological fluency task, where the participants had one minute to enunciate as many words beginning with a certain phoneme as they were able to, after being instructed to avoid repetitions. The participants could say words belonging to any part of speech, e.g., proper nouns, common nouns, verbs and adjectives. For example, for the phoneme /p/, the answers <Pedro>; <pau>, English translation <stick>; <presentear>, English translation <to give>; and <potente>, English translation <potent> are accepted.

After this, the phonological sentence task was applied. In this task, the assessor read the sentences without hesitation, and therefore this is another task that requires previous training. The participant then had to classify the sentences as correct or incorrect. Incorrect sentences are the ones containing the pseudowords. Additional tasks from the other domains were applied until the application of the DuLIP-EP was completed.

2.2.4. Data analysis

The data obtained from the 144 participants were initially recorded in an Excel database and subsequently analysed with IBM SPSS Statistics 24.

The stimuli adaptation hit rate was calculated per task and per item (number of correct items divided by the total number of items multiplied by 100). A cut-off score was defined to allow

only the items answered correctly by 90% of the sample to stay on the list. Items answered correctly by less than 90% of the general population were eliminated (Robert, Saul, & Arnold, 2007).

Descriptive statistics were used to characterise the sample and describe and summarise the data using different measurements. The sample was demographically characterised according to data available for the different regions of Portugal (INE, 2015). The handedness of the participants was also registered. Moreover, two gender groups, two age groups and two years of schooling groups were considered for the analyses of the results. The ranges used for the age groups mirrored those of the original study for two main reasons: 1) the average life expectancy is similar in the two countries (81.6 years in Portugal and 81.8 years in the Netherlands) (European Commission, 2017); and 2) although there is considerable discussion about the beginning of cognitive decline, several studies suggest that it starts at around 55 (Rönnlund, Nyberg, Bäckman, & Nilsson, 2005). The two groups created according to the years of schooling consisted of 12 years of schooling or less, and more than 12 years of schooling. The reasoning behind this choice was the current minimum mandatory years of schooling in Portugal (Decree-Law n.º 176/2012, Ministry of Education and Science, 2012).

The dependent variables included the number of correct answers per task. The phonological fluency variable was determined by calculating the average number of words said for the three phonemes of this task. The verbal diadochokinesis variable was determined by calculating the average number of seconds taken to pronounce the monosyllabic words in this task.

Graphic and statistical methods (Shapiro-Wilks test) were applied to explore the normality of the data distribution. Because the variables under consideration did not follow the normal distribution, the non-parametric Mann-Whitney U test was used to measure the effects of age, years of schooling and gender on ordinal test scores. Spearman's test was used to determine if there is a correlation between the phonological odd word task and the attention and memorisation domains.

Since the number of variables under study resulted in a need to reduce the statistical error of the tests, the p-value was corrected using the Šidák method (Šidák, 1967). A p-value of ≤0.0034 was used as the criterion of significance (5 x 3 for age, gender and education).

3. Results

3.1. Sample characterisation

The average age of the sample was 36.81±14.86 [age range 18 to 89] years old. Two age groups were established: 18 to 54 years old (n=121) and 55 years old and older (n=23). Both male (n=62) and female (n=82) participants were included. The average years of schooling were 15.36±4.14 [years of schooling, range 4 to 24]. Participants who integrated the >12 years of schooling group (n=132) outnumbered those from the ≤12 years of schooling group (n=32). According to the MMSE scores, none of the participants were cognitively impaired (28.64±0.97 points). Most of the participants were from Centre (n=94) and North (n=35) Portugal. The remaining belonged to the Lisbon Metropolitan Area (n=13) and the Autonomous Region of Madeira (n=2). The demographic data of the participants are detailed in Table 3.

Table 3: Demographic characteristics of the participants

Demographics	AVG	SD	Range
Age (y)	36.81	14.86	18-89
Schooling (y)	15.36	4.14	4-24
MMSE	28.64	0.97	25-30
Demographics	**Groups**	**Number of participants**	**Percentage**
Gender	M	62	43.10%
	F	82	56.90%
Age (y)	18-54 y	121	84.03%
	+55 y	23	15.97%
Education	≤ 12	32	22.22%
level	>12	112	77.78%
Region	North	35	24.31%
	Centre	94	65.28%
	Lisbon Metropolitan Area	13	9.03%
	Autonomous Region of Madeira	2	1.40%
Handedness	L	3	2.08%
	A	1	0.67%
	R	140	97.22%

Legend: AVG = average; SD = standard deviation; MMES = Mini Mental Examination State; M = male; F = female; y = years; L = left-handed; A = ambidexter; R = right-handed

3.2. Adaptation results

In the adaptation of the word repetition task, 74% of the items, corresponding to 222/300, were changed, since the literal translation did not meet the linguistic criteria specified in the original DuLIP. These changes were either new insertions or transfers from one level to another. A total of 140 words (47%) were inserted and 82 (27%) were moved from their original group and integrated into another complexity group. The remaining 78 words (26%) resulted from literal translations and were not moved into another complexity level. Details on the number of items changed by complexity level can be found in Table 4. The hit rate for this task was 99.63%. However, this was calculated on a per item basis. Only the the word <limiar> had a low hit rate (78.47%), which was lower than the cut-off score previously defined (90%). Therefore, this item had to be eliminated. After it was eliminated, the hit rate was recalculated and a score of 99.70% was achieved for the word repetition task.

Table 4: Items changed in the word repetition task

Linguistic level	Task	Complexity level	Adaptation of stimuli			
			O.T.	C.C.L.	N.W.	Total
Phonology	Repetition of words	1	23	35	17	52/75
		2	8	4	38	42/50
		3	7	19	14	33/40
		4	2	19	14	33/40
		5	36	14	50	64/100
		6	2	6	17	23/25

Legend: O.T. = Original translation; C.C.L. = Changed complexity level N.W. = New words

The phonological odd word out task included 60 monosyllabic words distributed across 15 groups. The items inside each group shared the same syllabic structure. The CVG structure represented 40% of the groups; the CVC structure represented 33.3%; the CV structure represented 20%; and CVGC structure represented 6.7%.

The hit rate initially calculated for this task was 96.13%. After removing item 8, which had a hit rate of 88.89%, a final score of 96.69% was reached. This item consists of the sequence '<chá-pó- lá-pá> ([ˈʃaˈpɔ ˈlaˈpa]), translation of <tea-dust-there-shovel>.

The hit rate for the phonological judgment task was 99.75%. None of the items scored less than 90%, so no sentences were deleted. In the verbal diadochokinesis task, 5 items exceeded the 4-second mark.

No analysis of the hit rate and cut-off point was carried out for the articulation and phonological fluency tasks. Those variables were only analysed through descriptive statistics (see section 3.3.). Table 5 outlines the hit rate for each task and the items that had to be eliminated.

Table 5: Adaptation results

Linguistic level	Tasks	Hit rate (initial-final)	Items eliminated (%)
Phonology	Word repetition	99.63% - 99.70%	Limiar (78.47%)
	Phonological odd word out	96.13% - 96.69%	Item 8 (88.89%)
	Phonological judgment	99.75%	----------------------------
	Phonological fluency	----------------------------	----------------------------
Articulatory	Verbal diadochokinesis	----------------------------	----------------------------

3.3. Standardisation results

No significant differences were found between the male and female participants of this study, both in terms of average age (p = 0.269) and average years of schooling (p = 0.293). In general, the participants obtained high scores, meaning that the average score is not far behind the maximum score.

Similarly to the original study, the average, median, ranges and cut-off scores were calculated for each linguistic task. Percentiles 2 and 7 were used as reference points. This method is commonly accepted in clinical practice as a way to set the clinical impairment threshold (Palmer, Boone, Lesser, & Wohl, 1998). Each of these results was analysed not only globally but also according to various age/education combinations. Consequently, Group 1 comprised the younger participants (18-54 y) with the least years of schooling (≤12 y), Group 2 comprised the older participants (>54 y) with the least years of schooling (≤12 y), Group 3 comprised the younger participants (18-54 y) with the most years of schooling (>12 y) and Group 4 comprised the older individuals (>54 y) with the most years of schooling (>12 y). The results obtained are presented in Table 6. With the exception of Group 3 (n=102), the remaining Groups (1, 2 and 4) had a very low sample size (n=19; n=13; n=10, respectively) and no percentiles were set for them.

Table 6: Descriptive statistics for phonological and articulatory tasks of DuLIP-EP

Linguistic level	Parameters	Group 1	Group 2	Group 3	Group 4	Total
Phonology (maximum score)						
Repetition (299)	AVG.	297.63	297.69	298.25	298.30	298.13
	Median	298.00	298.00	299.00	299.00	299.00
	Range	[294-299]	[295-299]	[295-299]	[297-299]	[294-299]
	P2	---------	---------	296.00	---------	295.00
	P7	---------	---------	297.00	---------	296.00
Phonological odd word out (14)	AVG.	13.47	13.00	13.58	13.14	13.35
	Median	14.00	14.00	14.00	14.00	14.00
	Range	[11-14]	[10-14]	[4-14]	[9-14]	[4-14]
	P2	---------	---------	7.12	---------	8.80
	P7	---------	---------	13.00	---------	12.00
Phonological sentence judgment (n=30)	AVG.	29.95	29.85	29.95	30.00	29.94
	Median	30.00	30.00	30.00	30.00	30.00
	Range	[29-30]	[29-30]	[28-30]	[30-30]	[28-30]
	P2	---------	---------	29.00	---------	29.00
	P7	---------	---------	30.00	---------	30.00
Phonological fluency (no max)	AVG.	16.00	13.08	18.44	18.80	17.66
	Median	15.33	14.00	17.67	17.33	17
	Range	[9.67-22.67]	[7-20.33]	[9.67-33]	[13.67-29]	[7-33]
	P2	---------	---------	10.35		8.33
	P7	---------	---------	11.33		11.00
Articulatory (time in seconds)						
Verbal diadochokinesis	AVG.	2,56	3.64	2.92	3.05	2.95
	Median	2,48	3.03	2.73	2.90	2.73
	Range	[1,92-4,63]	[1.99-6.90]	[1.74-5.85]	[2.31-4.28]	[1.74-6.90]
	P2	---------	---------	1.93	---------	1.92
	P7	---------	---------	2.09	---------	2.09

The analysis of the results of the Mann-Whitney Test (Table 7) showed that, for a p-value=0.05, most of the tasks used were not affected by gender/age/years of schooling. However, in the phonological domain, it was possible to observe that the word repetition task was impacted by the gender (p=.002), with a better performance in the female group. Additionally, the phonological odd word out task was impacted by the years of schooling (p=.010), with a higher average score in the group with the most years of schooling (>12 years of schooling). The average scores in the fluency task varied significantly according to the years of schooling (p=<.001), age (p=.020) and gender (p=.005). The younger participants, the participants with the most years of schooling and the female group achieved higher scores. No significant differences were found in the phonological judgment task and the articulation task.

When using a p-value≤0.0034 as the criterion of significance, the repetition task was impacted by the gender and the fluency task was only impacted by the age and years of schooling.

Table 7: P-values per category and task

DuLIP-EP task (maximum score)	Age	Years of schooling	Gender
Word repetition (n = 299); MWU	U = 1304.50, Z = -.522, p = .602	U = 1468.50 Z = -1.709, p = .087	U = 1845.50, Z = -3.089, p = .002
Phonological odd word out (); MWU	U = 1209.50, Z = -1.299, p = .194	U = 1381.00, Z = -2.585, p = .010	U = 2382.00, Z = -.845, p = .398
Phonological fluency (no maximum score); MWU	U = 964.00, Z = -2.332, p = .020	U = 1025.00, Z = -3.687, p < .001	U = 1844.5, Z = -2.815, p = .005
Phonological sentence judgment (30); MWU	U = 1329.00, Z = -.915, p = .360	U = 1689.50, Z = -1.322, p = .186	U = 2469.50, Z = -.785, p = .432
Verbal diadochokinesis; MWU	U = 1059.50, Z = -1.811, p = .070	U = 1767.50, Z = -.118, p = .906	U = 2169.00, Z = -1.505, p = .132

Calculations were made to determine whether there was a correlation between the memory and attention components and the phonological odd word out task in this study. These calculations showed a correlation coefficient between the memorization scores of the ACE-III and the analysed task of r=0.149 and an associated significance level of sig=0.155. In terms of the attention scores of the ACE-III, the correlation coefficient was r=0.149 and the significance level was sig=0.074. In both cases, the relation between the variables was very low and not statistically significant (Hemphill, 2003).

4. Discussion

This study describes the linguistic and cultural adaptation of the DuLIP for use in EP. It also gives a detailed account of the contribution made towards standardising this test battery, which was designed for use under specific awake brain surgery conditions. What follows is a discussion of the data obtained. Limitations of this study and future research guidelines are proposed in the last chapter of this Thesis.

The quantitative analysis of the data showed a ceiling effect for almost all tasks. The exceptions were the fluency and diadochokinesis tasks, in which there was no maximum score. The majority of the tasks present in the original study to evaluate the phonological and articulatory domains were kept in the adaptation of the DuLIP to EP. The only exception was the phonological odd word out task, which was eliminated from the original study but kept in the EP version. The goals of each specific task were also maintained. During the adaptation process, an effort was made to keep the maximum possible number of original DuLIP items, using their literal translation. Nevertheless, all of these items' scores had to display specific psycholinguistic properties. Accordingly, some items had to be eliminated or replaced due to linguistic and cultural incompatibilities or because they had a hit rate of less than 90% when the DuLIP-EP was applied.

In the word repetition task, some words (literal translations) did not fit into any other levels because the number of syllables varied significantly between Dutch and Portuguese. These words were therefore eliminated. For example, the word <trouwerij>, whose English translation is <wedding> and whose Portuguese translation is <casamento>, could not be transferred to another complexity level because it has 4 syllables (*ca-sa-men-to*) and the word repetition task does not include polysyllabic words. All the combinations represented in the EP syllabic inventory (V, CV, CCV, VC, CVC and CCVC) were considered in the adaptation of this task (Mateus & D'Andrade, 2000). Furthermore, the most frequent consonant clusters in EP, i.e. plosive + liquid, fricative + liquid (Vigário & Falé, 1993) were contemplated. These are [pr], [br], [tr], [dr], [kr], [gr], [pl], [bl], [kl] and [gl] in the beginning or middle of a word; [tl] in the middle of a word; [fr] and [fl] in the beginning or middle of a word; and [vr] in the middle of a word (Instituto Camões, 2006; Mateus et al., 2005).

The word repetition task (AVG=298.13; SD=1.15) has a maximum score of 299. The word <limiar> was eliminated from this task. This word was repeated incorrectly by 31 participants, with 30 participants (43.2%) changing it to <linear>. One of the reasons behind this misinterpretation might have been the auditory similarity of the words, [limiˈar] and [liniˈar], with a single different phoneme (/m/ vs /n/). Existing acoustic evidence shows that these phonemes have a similarly low first formant, located in the 200-250 Hz range in an adult (Fant, 1970; Johnson, 2012).

In the phonological odd word out task (AVG=13.35; SD=1.306), item 8 was eliminated because it had a hit rate inferior to 90%. Like many other tasks in the DuLIP, this one required a high degree of attention and memorisation, since the words were spelt sequentially and the participant had to memorise the words until the last one so that they could then identify the

intruder. When ascertaining whether there was a correlation between the scores obtained in the attention (AVG=17.30; SD= 1.052) and memorisation (AVG= 24.17; SD= 2.455) domains and the results in this task, there was a low correlation with no statistical significance. With this in mind, it should be remembered that the repetition of the auditory stimuli was allowed in the DuLIP whenever the participants so requested. During the application of this task, it was informally noticed that many participants asked for a repetition of the stimuli. When repeating the stimuli, the attention and memorisation abilities of the participants were reinforced (Haubrich & Nader, 2018; Tulving & Madigan, 1970). For this reason, the correlation obtained can be biased. This task was scored with either a 0 or a 1, like the other tasks. However, it could have been relevant to define three different scores: 0 points for not answering or answering incorrectly; 1 point for answering correctly after requesting a repetition; 2 points for answering correctly without requesting a repetition.

In the phonological fluency task (AVG=17.66; SD=5.011), the years of schooling were the only variable with influence when a corrected p-value was used as the criterion of significance. Similar conclusions were found in the literature, which mentions that the effects of age and gender on phonemic fluency are less pronounced than the effects of the years of schooling (Cavaco et al., 2013).

The diadochokinesis task of the DuLIP has 9 items. However, in the application of the DuLIP-EP, a high number of items took more than 4 seconds to complete. Because this task is performed during DES, these items had to be eliminated to ensure that the instrument is compliant with the specific conditions of the awake brain surgery. The items eliminated were [pafpafpaf], [paftafkaf], [dafnaflaf], [pafpofpuf] and [pafpaʃpag], which have a CVC syllabic structure. In EP, the /f/ and /g/ phonemes never occur at the end of a syllable (Jesus et al., 2015; Mateus et al., 2005). In the sequences presented, only the /ʃ/ of /paʃ/ can occur at the end of a syllable. Nevertheless, this phoneme is located in the middle of two syllabic structures that contain phonemes at the end of the syllable, which cannot occur in EP. This can influence the performance of the participants, as stated in other studies, which point out that the linguistic nature of the stimuli can have an impact on the verbal diadochokinesis rate (Scott Yaruss & Logan, 2002). A significant variation in the rate of syllabic production was found between languages, suggesting that language may be an important factor. It seems that an oral-DDK rate does not only represent a physiological ability, but also a culturally-bound trait (Icht & Ben-David, 2014). After eliminating these items, the analysis of the results showed an AVG=2.95; SD=0.878.

In the quantitative analysis of the data, the average scores obtained in the DuLIP and DuLIP-EP were not compared because two items had to be eliminated in the Portuguese version, leading to a different number of total items. However, some differences regarding the gender, age and years of schooling were found.

In the DuLIP, the gender of the participants did not have a measurable impact in any of the tasks. However, performance differed significantly according to the age and the years of schooling. An exception to this was the verbal diadochokinesis task, which wasn't measurably impacted by any of these. The phonological odd word out task was not measurably influenced by the age. In comparison, in the DuLIP-EP, the years of schooling influenced the fluency task and the gender

influenced the repetition task. No significant differences were observed between age/gender/years of schooling in the phonological sentence judgment task and in the verbal diadochokinesis task, as in the original study. In terms of the number of participants placed into each group according to their age and years of schooling, the original study was more balanced than this one, which might have had some influence on the results.

5. Conclusions

The main goal of this study was to linguistically and culturally adapt the phonological and articulatory tasks of the DuLIP, as well as to contribute to their standardisation in EP. The test battery has a wide range of language levels, but this study focused only on the phonological and articulatory components. Accordingly, stimuli were carefully created based on literal translations and in compliance with the psycholinguistic properties of the original study. The test battery was then applied to a sample of 144 healthy Portuguese participants.

Data were analysed according to the age, gender and years of schooling of the participants. The results show that the tasks have a high hit rate and their average scores are close to the maximum possible.

This study allowed the development of a test battery that assesses language and articulation skills in pre-, intra- and postoperative moments for EP. The DuLIP-EP has log sheets with brief and succinct application instructions.

This test battery has yet to be validated. However, as a suggestion, the final version of this study could be applied in the development of other projects. To the author's best knowledge, no other language evaluation protocol for use during awake brain surgery has yet been validated or adapted into EP. For that reason, this is an innovative study in Portugal, where it will fill an existing gap in clinical practice and contribute to improving the quality of life of patients.

5.1. Future work

Although there was a reasonable number of participants, it would have been relevant to broaden this study to even more people and thus have more balanced groups, especially the ones with the youngest participants and lowest years of schooling. It could also be relevant to cover more regions of Portugal. Additionally, it could be pertinent to create an application manual with detailed instructions on procedures, scoring methods and thresholds for clinical impairments. Regarding the adaptation of the stimuli, future work could use distinctive features to analyse the items in the phonological tasks.

The validation of the DuLIP-EP in clinical practice would be valuable. Subsequently, a longitudinal study on the postoperative recovery of the patient, as well as on their quality of life, would be equally relevant. After all, language is the key to communication and plays a major role in quality of life.

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Scientific outputs developed under this master's degree

Work published under this master's degree

Jesus, L., Alves, J., Cardoso, M., & Morgado, M. (2019). Linguistic Intraoperative Protocol. In *Proceedings ExLing 2019: 10th International Conference of Experimental Linguistics* (pp. 121-125). Lisboa, Portugal: ExLing Society.

Oral presentations under this master's degree

Oral communication presented at the 10th International Conference of Experimental Linguistics

Conference attendance grant

Grant from the Organization Committee of the 10th International Conference of Experimental Linguistics

Appendices

Appendix I: Participants' informed consent

Consentimento Informado, Livre e Esclarecido para Participação em Investigação de acordo com a Declaração de Helsínquia

(Helsínquia 1964; Tóquio 1975; Veneza 1983; Hong Kong 1989; Somerset West 1996; Edimburgo 2000; Washington 2002; Tóquio 2004; Seoul 2008)

Título do estudo: Tradução, Adaptação e Validação para Portugal do *Dutch Linguistic Intraoperative Brain Protocol (DuLIP).*

Enquadramento: Este estudo enquadra-se no Mestrado em Terapia da Fala, da Escola Superior de Saúde da Universidade de Aveiro orientado pelo Professor Doutor Luís Jesus e visa traduzir e adaptar um instrumento de avaliação da linguagem em contexto cirúrgico e pós-cirúrgico, mais especificamente das áreas semântica, fonológica e morfossintática. Avalia ainda a articulação verbal. A sua colaboração contribuirá para a adaptação de um teste inovador para o português europeu e, posteriormente, para a melhor compreensão do mapeamento cerebral no que concerne às áreas da linguagem e fala.

Explicação do estudo: Este é um estudo científico que envolve entrevistas e aplicação de testes em local e horário que lhe sejam convenientes. Será solicitada a resposta a determinadas questões demográficas e clínicas. Além disso, será pedida a resposta a determinados itens específicos que o teste a validar contempla.

O Dutch Linguistic Intraoperative Protocol (DuLIP) é um instrumento de origem Holandesa, criado e validado para a avaliação de competências de linguagem em contexto pré, intra e pós cirúrgico em pacientes diagnosticados com lesões tumorais a nível cerebral e submetidos a estimulação elétrica direta (DES) – uma técnica utilizada em neurocirurgia com o paciente acordado, que permite o mapeamento das regiões cerebrais corticais e subcorticais. Embora esta técnica seja cada vez mais utilizada e já considerada um procedimento *gold standard* em neurocirurgia, ainda carece de instrumentos validados para a avaliação das competências a testar no aparato cirúrgico.

No que refere às lesões tumorais que afetam as áreas da linguagem, sabe-se que existem benefícios na utilização desta técnica relativamente ao *outcome* linguístico pós-operatório. Não obstante, a existência de métodos de identificação das áreas associadas a estas competências, validados para os contextos anteriormente referidos são escassos e, no caso do Português Europeu, de acordo com a pesquisa bibliográfica realizada, inexistentes. Assim, este estudo poderá contribuir para a prática clínica das áreas de Neurocirurgia, Neurologia, Terapia da Fala, Neurolinguística e Neuropsicologia, maximizando os resultados e possível potencial de reabilitação dos utentes. Permitirá ainda contribuir para o melhor mapeamento de regiões e trajetórias cerebrais, corticais e subcorticais, relacionadas com a linguagem, nomeadamente nas áreas da fonologia, semântica e sintaxe. As capacidades articulatórias também são contempladas no instrumento.

Condições de financiamento: A participação no estudo não é remunerada. A decisão de participar no mesmo é inteiramente sua. Caso aceite participar, poderá desistir a qualquer momento, bem como recusar qualquer procedimento, sem ser penalizado por isso.

Possíveis riscos ou desconforto: As tarefas não apresentam quaisquer riscos ou desconforto para os participantes.

Confidencialidade e anonimato: A confidencialidade dos dados e dos envolvidos no estudo está salvaguardada. A identificação pessoal ou dados alusivos à mesma não serão disponibilizados a outro grupo ou instituição.

Gratas pela disponibilidade demonstrada, encontramo-nos disponíveis para esclarecer qualquer questão relativa ao estudo.

Contacto para possível esclarecimento de dúvidas:

Joana Alves	Mafalda Cardoso	Mariana Morgado
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Este documento é composto de 2 páginas e feito em duplicado: uma via para o/a investigador/a e outra para a pessoa que consente

DECLARAÇÃO DE CONSENTIMENTO

de acordo com a Declaração de Helsínquia

(Helsínquia 1964; Tóquio 1975; Veneza 1983; Hong Kong 1989; Somerset West 1996; Edimburgo 2000; Washington 2002; Tóquio 2004; Seoul 2008)

Título do estudo: Tradução, Adaptação e Validação para Portugal do *Dutch Linguistic Intraoperative Brain Protocol* (DuLIP).

Eu, ____, abaixo-assinado, declaro ter lido e compreendido este documento, bem como as informações orais que me foram fornecidas pela(s) pessoa(s) que assinaram abaixo.

Foi-me dada a oportunidade de fazer as perguntas que julguei necessárias, e a todas obtive resposta satisfatória.

Tomei conhecimento de que, de acordo com as recomendações da Declaração de Helsínquia, a informação ou explicação que me foi prestada versou os objetivos, os métodos, os benefícios previstos, os riscos potenciais e o eventual desconforto. Além disso, foi-me garantida a possibilidade de, a qualquer momento, recusar participar no estudo sem qualquer tipo de comprometimento.

Eu compreendo que os resultados do estudo podem ser publicados em revistas científicas, apresentados em conferências e usados noutras investigações, sem que haja qualquer quebra de confidencialidade. Portanto, dou autorização para a utilização dos dados para esses fins. Assim, aceito participar neste estudo e permito que a utilização dos dados que de forma voluntária forneço, confiando que são utilizados confinadamente para o que me foi descrito e me é garantida, pelos investigadores, a confidencialidade e anonimato dos mesmos.

P'los investigadores, O participante,

____________________________________ ____________________________________

_________, ___ de ___________________ de 2019

Este documento é composto de 2 páginas e feito em duplicado: uma via para o/a investigador/a e outra para a pessoa que consente

Appendix II: Anamnesis

CÓDIGO___________

PROTOCOLO PARA RECOLHA DE DADOS

Data da realização do teste: ___/___/_____

Dados recolhidos por: ________________

IDENTIFICAÇÃO PESSOAL

Região de residência: ____

Sexo: Masculino (__) Feminino (__)

Data de nascimento: ___/___/_____ Idade ____ anos

FORMULÁRIO DE INFORMAÇÃO PESSOAL

Lateralidade: Destro (__) Esquerdino (__)

Grau de escolaridade: Não sabe ler nem escrever (__) Sabe ler e escrever (__)

Ensino primário (__) 2º ciclo (__) 3º ciclo (__) Ensino secundário (__)

Pós-graduação (__) Licenciatura (__) Mestrado (__) Doutoramento (__)

Indicar o número de anos de estudos: ____

Situação de empregabilidade atual: Estudante (__) Empregado (__) Desempregado (__) Reformado (__)
Profissão ou antiga profissão: ____

Informação linguística

Língua materna: Português (__) Outro (__)

Qual a língua que fala em casa? Português (__) Outro (__) Especifique: ___________________________

Qual a língua na qual recebeu educação? Português (__) Outro (__) Especifique: __________________

É fluente noutras línguas? Português (__) Inglês (__) Francês (__) Espanhol (__) Especifique: __________

História clínica

Antecedentes neurológicos (e.g. epilepsia)? Sim (__) Não (__) Especifique: __________________________

Antecedentes psiquiátricos? Sim (__) Não (__) Especifique: ______________________________________

Alterações de aprendizagem ou comportamentais? Sim (__) Não (__) Especifique: ___________________

Alterações de fala e/ou linguagem? Sim (__) Não (__) Especifique: ________________________________

Alterações relacionadas com visão/perceção de cores? Sim (__) Não (__) Corrigida (___)

Especifique: ____

Alterações relacionadas com audição? Sim (__) Não (__) Corrigida (__) Especifique: __________________

Dependente de substâncias tóxicas álcool ou drogas? Sim (__) Não (__) Especifique: _________________

Medicação que possa influenciar os resultados (e.g. medicação indutora de sono; psicofármacos; medicação neuroléptica)? Sim (__) Não (__) Especifique: ____

Antecedentes cardiovasculares: Sim (__) Não (__)

Observações (e.g. observação comportamental durante o teste):

____

____

____

Appendix III: Ethical Approval

COMISSAO DE ETICA

da **Unidade Investigação em Ciências da Saúde: Enfermagem** (UICISA: E)

da **Escola Superior de Enfermagem de Coimbra** (ESEnfC)

Parecer Nº 545/ 01-2019

Título do Projecto: Tradução, Adaptação e Validação para Portugal do Dutch Linguistic Intraoperative Brain Protocol (DuLIP)

Identificação das Proponentes

Nome(s): Joana Patrícia Gomes Alves, Mafalda Inês Martins Cardoso e Mariana Morgado Oliveira Martins

Filiação Institucional: Escola Superior de Saúde da Universidade de Aveiro (alunas de mestrado em terapia da fala)

Investigador Responsável/Orientador: Prof. Luís Miguel Teixeira de Jesus

Relator: Sofia Raquel Teixeira Nunes

Parecer

Considerando a avaliação das competências linguísticas pré, intra e pós-operatórias em doentes com lesões tumorais a nível cerebral e submetidos a estimulação elétrica cerebral, este estudo tem como objetivo traduzir e adaptar ao português europeu o instrumento DuLIP e validar o referido à população portuguesa normal. Segundo os investigadores, será um estudo do tipo metodológico com fase qualitativa e quantitativa (a primeira respeita a tradução e adaptação do instrumento e a segunda respeita à aplicação do mesmo).

A data de início de colheita de dados encontra-se prevista para 1 de fevereiro de 2019 e o término a 1 de março de 2022, pelo que à data atual terão os investigadores de adaptar as mesmas.

A amostra agregará no mínimo 140 pessoas da população portuguesa, com critérios de inclusão bem definidos. Será realizada uma amostragem por conveniência em diversas regiões de Portugal.

Os investigadores irão utilizar para além dos instrumentos de colheita de dados, instrumentos que permitam aferir da compatibilidade com os critérios de inclusão dos participantes.

Referiram os investigadores que toda a informação recolhida será tratada de forma confidencial, onde são garantidas a voluntariedade e a autonomia dos participantes. Os dados serão utilizados mas os nomes dos participantes serão substituídos por códigos e só os investigadores terão acesso aos dados. Contudo, é referido que irão recolher o nome dos participantes mas que estarão em destacáveis que serão retirados. Foram verificadas algumas situações na identificação dos utentes e no folha informativa que prontamente foram corrigidas pelos investigadores.

Sendo assim, somos do parecer que o projeto pode ser aprovado sem restrições de natureza ética.

O relator:

Data: 12/03/2019 O Presidente da Comissão de Ética: ____________