



Università degli Studi di Padova
Dipartimento di Medicina Molecolare

Corso di Laurea Magistrale in
Scienze Riabilitative delle Professioni Sanitarie
Presidente: Ch.mo Prof. Daniele Rodriguez

TESI DI LAUREA

VALIDATION OF A TEST TO ASSESS DYSARTHRIA IN
NEUROLOGIC PATIENTS: A CROSS-SECTIONAL PILOT STUDY

RELATORE: Prof.ssa Frigo Anna Chiara

Correlatore: Dott. Turolla Andrea

LAUREANDO: Dott. De Biagi Francesca

Anno Accademico 2015-2016

Table of contents

| | |
|--|----|
| <i>Abstract</i> | 3 |
| <i>Introduction</i> | 4 |
| <i>Background</i> | 5 |
| Dysarthria definition | 5 |
| Dysarthria classification..... | 6 |
| Dysarthria epidemiology..... | 8 |
| Dysarthria assessment | 10 |
| <i>Aim</i> | 12 |
| <i>Method</i> | 13 |
| Study design | 13 |
| Subjects | 13 |
| Scorers..... | 16 |
| Procedure..... | 16 |
| <i>Materials</i> | 18 |
| <i>Results</i> | 20 |
| Administration..... | 20 |
| Offline scoring: intra-rater and inter-rater reliability | 20 |
| Online scoring: inter-rater reliability | 26 |
| <i>Discussion</i> | 27 |
| <i>Psychometric features</i> | 27 |
| Clinical utility..... | 29 |
| Limitations | 30 |
| <i>Conclusions</i> | 31 |
| <i>References</i> | 33 |
| <i>Acknowledgments</i> | 38 |
| <i>Enclosed</i> | 39 |

Abstract

Dysarthria is a motor speech disorder that results from an impairment of the muscles devoted to speech production, thus affecting the movements of the orofacial district. The type and severity of dysarthria depend on which structures of the central or peripheral nervous system are affected. Due to the vast range of neurological disorders that may cause dysarthria, its prevalence may be not negligible.

The aim of this study was to explore the reliability of a short-form of a protocol to assess dysarthria, which is broadly used in the Italian clinical practice and that was standardized but never validated.

Fifty dysarthric patients were enrolled for this pilot study and assessed by the protocol ("*Protocollo di Valutazione della Disartria*"; Fussi, Cantagallo, Bertozzini, 1997; revised by De Biagi *et al.* 2015). We determined the consistency of measurements between the same rater and among different raters with different level of expertise by the Lin's Concordance Correlation Coefficient (CCC). Scores were given both online, within the assessment, and offline, watching the video recordings of patients' evaluations.

Preliminary results indicated an excellent consistency of ratings in repeated measures over time (offline intra-rater CCC > 0.8). Nevertheless, it was shown a slight stability of ratings among different scorers (offline inter-rater CCC < 0.8), especially in the online administration of the protocol (online/offline inter-rater CCC < 0.8). The protocol showed its potential clinical utility due to its completeness as well as its facility of administration.

Although the protocol seemed to be a potentially useful test, generalizations of the findings are limited, due to the characteristics of the study, which was monocentric and with a small sample size. Indeed, further research is required for a better validation of the instrument.

Introduction

Neurological injuries often impair many functions underpinning the whole process of communication. Dysarthria is one of the communicative impairments whose etiopathology may be attributed to vast range of both acute and degenerative neurological illnesses.

The term dysarthria refers to an altered speech production resulting from a neurological injury involving the motor component of speech process. Although there are no data regarding its prevalence within the general population, the disease is not a rare condition. Moreover, dysarthria might be one of the most disabling conditions resulting from a neurologic disease as it affects communicative ability, therefore it may have a strong impact on many relevant aspects, such as employment, personal and social life. Communicative disabilities may in fact reduce participation as defined by the World Health Organisation's (W.H.O.) Classification of Functioning Disability and Health (I.C.F., 2001) and may lead to isolation, depression and loss of independence.

For these reasons, a standardized and validated tool to assess dysarthria is required to plan the right goals of the treatment, but mostly to measure the effectiveness of rehabilitation programs.

Accordingly, the purpose of this study is to measure the reliability of a short form of a test to assess dysarthria (*"Protocollo di Valutazione della Disartria"*; Fussi, Cantagallo, Bertozzini, 1997; revised by De Biagi *et al.* 2015), which is broadly used in Italy in clinical practice.

Background

Dysarthria definition

Many speech and language therapists (SLT) and neurologists, still generally accept the following definition of dysarthria given by Aronson, Darley and Brown: “*Dysarthria is a collective name for a group of speech disorders resulting from disturbances in muscular control over the speech mechanism due to a damage of the central or peripheral nervous system. It designates problems in oral communication due to paralysis, weakness, or incoordination of the speech musculature.*” (Aronson, Darley & Brown, 1969)

Accordingly, this definition implies that dysarthria (Duffy, 2013):

- is due to a neurological disease, affecting central and/or peripheral nervous system;
- is attributed to an abnormal muscular activation of the structures involved in speech production, including lips, tongue, vocal folds and soft palate;
- may be extremely variable in terms of severity and characteristics;
- should be distinguished from other language disorders (e.g. aphasia) or cognitive impairments (e.g. dementia). Furthermore, dysarthria does not originate from anatomic structures abnormalities (e.g. cleft palate), sensitivity loss (i.e. deafness) or psychological problems. Moreover, the term does not refer to apraxia of speech, defined as the “Neurologic speech disorders reflecting an impaired capacity to plan or program sensorimotor commands necessary for directing movements that result in phonetically and prosodically normal speech” (Duffy, 2005).

Thus, a person with dysarthria may demonstrate the following characteristics:

- altered breathing mechanisms;
- "slurred," "choppy," or "mumbled" speech that may be difficult to comprehend;
- inaccurate speech production, leading to phoneme distortions;
- slow or excessive rate of speech;
- irregular pitch and rhythm when speaking;
- changes in voice quality, such as hoarse or breathy voice or speech that sounds "nasal" or "stuffy".

Dysarthria classification

Motor speech disorders vary across different dimensions so that many classification systems have been proposed in the last years. Dysarthria could be classified taking into account different criteria (Pezzella *et al.*, 2013). One of the variables that may be considered is the age at onset; in fact, dysarthria can be either acquired or congenital. Even if this distinction is fundamental for clinicians to manage these disorders, this thesis will be focused on acquired ones. The course of the illness is also an important variable. Dysarthria may be classified as congenital, chronic or stationary, improving and progressive or degenerative. Monitoring dysarthria over time could help also to control the course of the underpinning disease. Other classifications are based on the site of lesion. Dysarthria may result from lesions in different body structures, such as: neuromuscular junction, peripheral and cranial nerves, brainstem, cerebellum, pyramidal and extra pyramidal pathways and left or right cerebral cortex. Knowledge of the exact site impaired may supply important information about the expected speech disorders or even other significant comorbidities (i.e. cognitive and/or motor disabilities).

Other distinctions are based on the neurologic diagnosis; a vast range of neurologic impairments may in fact cause dysarthria, including vascular, inflammatory, degenerative, neoplastic, toxic, metabolic, traumatic and even developmental aetiologies. Even if the acquaintance of the aetiology is fundamental, it is neither valid, nor feasible to classify motor speech disturbances by the underpinning impairments, as it may effect multiple and variable parts of the nervous system; for instance, it is not possible to identify and describe “the dysarthria of stroke”. Dysarthria may be classified considering the severity of the disorders; this construct appears inconsistent, as these kinds of speech disorders usually vary among a continuum between mild, average and severe.

The classification system that is still broadly accepted in literature is the *Mayo Clinic Classification System* (Darley, Aronson e Brown, 1969), which is based on the correspondence between the site of lesion (underlying pathophysiology) and the perceptual features of speech disorders (clinical manifestation).

The early version of this classification system distinguished six major types of dysarthria: spastic, ataxic, hypokinetic, hyperkinetic, flaccid and mixed. In 1975 the same authors added two other categories: unilateral upper motor and undefined dysarthria. Table 1 summarizes the characteristics of six types of dysarthria that may be appreciated most frequently in clinical practice.

| DYSARTHRIA TYPE | ATAXIC | SPASTIC | HYPOKINETIC | HYPERKINETIC | FLACCID | UNILATERAL MOTOR NEURON |
|----------------------------|--|--|--|--|---|---|
| SITE OF LESION | Cerebellar | Bilateral damage at upper motor neuron, pyramidal and extrapyramidal systems | Substantia nigra | Extrapyramidal tract, specifically basal ganglia | Peripheral or lower motor neuron system, neuromuscular junction | Unilateral lesion first neuromotor |
| NEUROLOGICAL DISORDERS | Cerebellar ataxia | Pseudobulbar palsy, | Parkinson Disease | Choreas and dystonias | Bulbar palsy, ALS | Stroke |
| PHYSIOLOGY | Inaccuracy of movement and Slowness of movement | Muscular weakness and spasticity | Slow muscular movements, limited range | Quick, unstained, involuntary movements | Weakness and lack of normal muscle tone | Muscular weakness and spasticity |
| ARTICULATION | Imprecise consonants or distortion | Imprecise consonants or distortion | Imprecise consonants or distortion | Imprecise consonants or distortion | Imprecise consonants or distortion | Imprecise consonants or distortion |
| PHONATION | Harsh, loudness may vary excessively | Strained-strangled or harsh voice quality, breaks | Hoarse or low volume | Abnormal | Breathiness of voice and nasal emission | Strained-strangled or harsh voice quality, breaks |
| PROSODY | Slow rate, prolonged phonemes and intervals, irregular articulatory breakdowns | Possible burst of loudness | Stoppages, monopitch, monoloudness | Voice stoppages | Slow rate and prolonged intervals, monopitch | Possible burst of loudness |
| RESONANCE | Tendency to place equal stress on syllables | Hypernasality | Hypernasality | Hypernasality | Hypernasality | Normal |
| SPEECH | Slurred, severely impaired | Slurred, sometimes described as explosive speech | Very slow or festinating | Varied across syndromes | Slow rate prolongation of sounds and intervals | Slurred |
| ASSOCIATED CHARACTERISTICS | Reduced facial expression | Reduced facial expression | Reduced facial expression, tremors | Reduced facial expression, emission of grunts noises | Insufficient respiratory support | Reduced facial expression |

Table 1. Clinical features of the six most frequent types of dysarthria

Dysarthria epidemiology

Dysarthria's aetiology may be attributed to many degenerative disorders, such as Parkinson's disease (PD) and Parkinsonism, Amyotrophic Lateral Sclerosis (ALS), progressive ataxias, Multiple Sclerosis (MS), myasthenia gravis. Moreover, dysarthria may be closely associated with many acute illnesses: cerebrovascular diseases, such as haemorrhages and strokes, Traumatic Brain Injuries (TBI), tumours, metabolic illness and infections. Furthermore, dysarthria may be caused by acute poisoning, or may result from neurosurgery.

Even if it is extremely difficult to appraise the exact prevalence and incidence of dysarthria within the general population, the disorder is not a rare condition (Hedge & Freed, 2011). In fact, it has been estimated that dysarthria may account for the 54% of all the acquired communication neurogenic disorders (Wang, 2010). Taking into account non-degenerative dysarthria, it has been estimated a prevalence of 60% of patients with traumatic brain injury during the acute stage of their recovery, and 10% at long term (Yorkston *et al.* 1999). Dysarthria is a frequent sign in cerebral ischemia ranging from 8% to 12.4% in large unselected stroke series (Urban *et al.* 2013), with an incidence up to 42% following first ischemic stroke (Flowers *et al.* 2013). Moreover, clinical trials frequently report the presence of dysarthria in both acute and subacute (3-months) phase after stroke (Ali *et al.* 2013). Regarding degenerative dysarthria, in a survey 70% of patients with PD indicated that their speech was impaired during the disease process (Hartelius *et al.* 1994) Dysarthria can appear at any stage of PD and worsens in the later stages of the disease to cause a progressive loss of communication and social isolation (Pinto *et al.* 2004). The prevalence of dysarthria associated with MS is 20% (Hartelius *et al.* 2000). Other studies indicate that 90% of people with moderately advanced ALS may present dysarthria. (Campbell & Enderby, 1984).

Although indisputable epidemiologic data are still lacking, dysarthria might be one of the most disabling outcome in association with a vast range of neurologic conditions. Dysarthria may have a severe impact on the quality of life of patients, which may often experience being laughed or ridiculed (Enderby, 2013). It has been highlighted that even mild dysarthria may have significant social and psychological effects (Mackenzie *et al.* 2014). For example, the possibility of not being able to communicate is one of the most distressing aspects of progressive neurological conditions. (Yorkston *et al.* 2007).

Dysarthria assessment

Generally, the aims of the motor function assessment of communication are similar to those for language impairments (Freed, 2012). The assessment should allow the detection of the primary problem in order to measure the baseline, thus to plan the proper goals of treatment (Haynes & Pinzola, 2011). At first, the examiner must gather relevant information on the history of the patient, who will then undergo to several examinations of the motor system function.

During the last decades, there has been a large diffusion of advanced technologies that allow the acoustical analysis of the verbal signal. Among them, the Multi Dimensional Voice Program (MDVP) by Key Pentax is a standard software that can calculate up to 33 different acoustic parameters from a voice sample, and which is widely used in the research field for being very comprehensive. (Christmann *et al.* 2015). Despite its potentials, this system is still not broadly used yet in clinical practice, partly because of the lack of a standard procedure and analysis (Nicastri *et al.* 2004).

In fact, the clinical assessment cannot exclude the direct examination of the patient (Schettino *et al.* 2013). Following, the main steps for the assessment of motor speech disorders are reported (Duffy, 2013):

- 1- Problem detection: features of speech should be accounted on the base of patient's history and description of the problem.
- 2- Differential diagnosis: when speech is abnormal, a list of diagnostic possibilities may be generated to make the differential diagnosis among motor speech impairments.
- 3- Dysarthria type diagnosis: once dysarthria has been detected, further examination should allow distinguishing among different dysarthria's type.

- 4- Disease characterization: speech should be described through oral mechanism examination, perceptual characteristics of speech and results of standard clinical tests.
- 5- Specifying severity: these esteem influences diagnosis and prognosis and will represent the baseline data to be compared with future changes.

The assessment procedures should comprehend: medical history, examination of speech structures, perceptual analysis of speech and judgment of intelligibility (Hedge & Freed, 2012).

Medical history reveals patient's observations of the disorder and gives relevant information on the underlying neurologic pathology, its onset and course and the associated symptoms.

The examination of speech structures should include the assessment of strength, speed, range, stability, tone and accuracy of muscular movements (Scettino *et al.* 2013). The non-verbal assessment should consider an examination of morphology, motility and sensitivity of the structures involved in speech production: respiratory muscles, larynx, lips, tongue, soft palate and jaw. The verbal assessment should comprise diadochokinetic tasks and stress testing.

The aim of the perceptual analysis is to assess the components involved in speech production: respiration, phonation, resonance, articulation and prosody. Assessment of intelligibility is crucial to set the right goals of treatment and it should be the main outcome measurement in all the cases of speech disorders (Bloch, 2011). First of all, it is necessary to differentiate between two types of intelligibility. The first is defined as signal-dependent intelligibility, which is the ability of the listener to understand the spoken message based solely on the sound signal. Whereas, contextual intelligibility is the understandability of the message due to the overall cues and clues from any other verbal (e.g. syntax, semantics) or non-verbal (e.g. facial expression, gesture,

broader contextual setting) sources. However, conflicting recommendations exist on how to measure it (Miller, 2013).

Many protocols have been developed to assess dysarthria. However, to our knowledge, only one tool allowing perceptual analysis of speech is available in Italian, that is “*Profilo di Valutazione della Disartria*” (Fussi, Cantagallo, Bertozzini, 1997), whose normative data have been provided with cross-cultural adaptation of “*Robertson Profile*” (Robertson, 1982).

This tool is divided into eight subscales (i.e. respiration, voice, facial musculature, diadochokinesis, reflex, articulation, intelligibility, prosody), each one including several items. Each item has a score ranging from 1 (worst) to 4 (best). The internal construct validity was investigated through a Rasch analysis in a sample of 196 patients (Cantagallo *et al.* 2006). The results of the study suggested the possibility of creating a short version of the test with a rescoring of the items in a 3-points scale.

Aim

The main aim of the study was to measure the reliability of a modified tool for the assessment of speech impairments (i.e. dysarthria) - “*Protocollo di Valutazione della Disartria*” (Fussi, Cantagallo, Bertozzini 1997; revised by De Biagi *et al.* 2015). In this regard, the following experimental hypotheses were tested:

1. Are inter- and intra-scorer reliability for the protocol adequate for clinical purposes?
2. Is the protocol a valid measure of speech production ability?

These preliminary data could be used to proceed with the first Italian validation of the protocol.

Method

Study design

The research design is an experimental cross-sectional pilot study for the validation of an outcome measure, aimed to assess dysarthria in patients with neurological diseases. The study was approved by the Ethic Committee of Venice on 31st May 2016 with the reference number 49A/CESC

Subjects

Fifty dysarthric patients (28 males and 22 females) volunteered to participate in this study. All patients hospitalized at IRCCS San Camillo Hospital Foundation (Lido di Venezia), diagnosed with dysarthria because of neurological etiology (August 2015 – May 2016) and referred by ward doctor for assessment of speech impairments.

The following inclusion criteria were considered for enrollment:

- ability to complete the protocol;
- consent to video recording;
- italian mother tongue.

Moreover, the following exclusion criteria were considered:

- inability to complete the protocol (e.g. severe cognitive disorders, aphasia, bucco-facial apraxia);
- open tracheotomy tube.

After enrolment, patients were divided into two groups, according to their diagnosis. Group 1 (N=25) was composed by patients diagnosed with degenerative dysarthria; group 2 (N=25) consisted of patients with non-degenerative dysarthria.

The subject's selection was conducted on purpose to reach the sample size of 50 subjects, which was fixed taking into account the rate of enrollment feasible for Speech and Language Therapy (SLT) service along the pre-determined duration of the study. These preliminary results will be used to estimate the adequate

sample size for future studies aimed to a better validation of the protocol.

In figure 1, the enrolment procedure is displayed. From August 2015 to May 2016 70 dysarthric patients were admitted to Hospital San Camillo and assessed by the SLT service. Six of them were excluded because the protocol was not feasible (among them 1 patient was diagnosed with aphasia, 3 patients suffered from bucco-facial apraxia and 2 other patients had severe cognitive disorders), 6 of them did not give the consent to video recording, 7 patients were not included due to technical problems and 1 was discharged before being assessed.

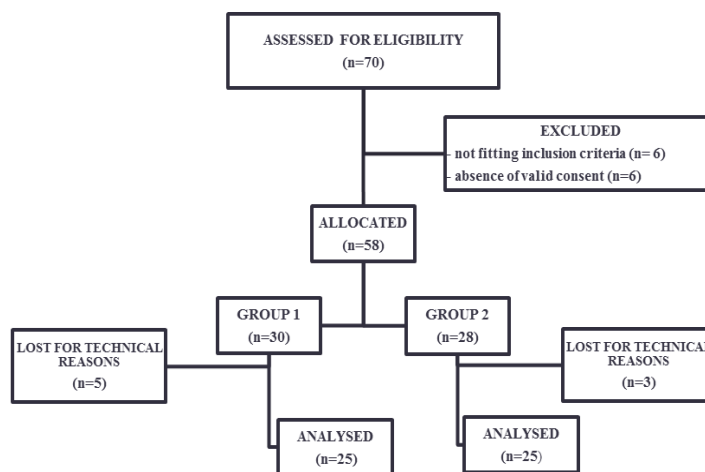


Fig. 1. Enrolment procedure

Table 2 summarizes descriptive statistic results of the 2 groups. Table 3 illustrates data of the included subjects.

| | GROUP 1 (N=25) | GROUP 2 (N=25) |
|-------------------------------|--------------------------------------|---|
| MEAN AGE | 58.48±9.96 | 63.36±11.10 |
| MEAN TIME POST ONSET (T.P.O.) | 145.32±126.16 | 62.4±152.97 |
| DIAGNOSIS | 7 MS, 7 PD, 5 ALS, 3 ataxia, 3 other | 3 left stroke, 10 right stroke, 2 Arnold-Chiari Syndrome, 3 TBI, 2 Subarachnoid hemorrhage, 5 other |

Table 2. Descriptive statistics of group 1 and group 2

| N | SEX | AGE (YEARS) | DIAGNOSIS | GROUP | T.P.O. (MONTHS) | DOMINANCE |
|----|-----|-------------|-----------|-------|-----------------|-----------|
| 1 | F | 40 | MS | 1 | 72 | R |
| 2 | M | 81 | LS | 2 | 0.5 | L |
| 3 | M | 49 | CA | 1 | 216 | L |
| 4 | M | 44 | ACS | 2 | 300 | L |
| 5 | M | 51 | ALS | 1 | 72 | L |
| 6 | F | 60 | ALS | 1 | 180 | L |
| 7 | M | 79 | PD | 1 | 108 | L |
| 8 | M | 72 | MS | 1 | 348 | R |
| 9 | F | 62 | ALS | 1 | 48 | L |
| 10 | M | 60 | MS | 1 | 84 | L |
| 11 | M | 61 | SH | 2 | 6 | R |
| 12 | F | 52 | MS | 1 | 408 | R |
| 13 | F | 50 | MS | 1 | 408 | R |
| 14 | M | 56 | LS | 2 | 1.5 | L |
| 15 | F | 58 | MS | 1 | 240 | L |
| 16 | M | 56 | TBI | 2 | 1.5 | R |
| 17 | M | 49 | PD | 1 | 60 | L |
| 18 | M | 65 | PD | 1 | 48 | L |
| 19 | M | 54 | ALS | 1 | 22 | L |
| 20 | F | 41 | MS | 1 | 312 | L |
| 21 | M | 73 | LS | 2 | 152 | L |
| 22 | F | 60 | PD | 1 | 36 | L |
| 23 | F | 61 | MSA | 1 | 60 | L |
| 24 | F | 71 | PD | 1 | 36 | L |
| 25 | M | 45 | PD | 1 | 96 | L |
| 26 | F | 70 | PD | 1 | 59 | L |
| 27 | F | 74 | CA | 1 | 36 | L |
| 28 | M | 61 | PD | 1 | 120 | R |
| 29 | F | 73 | SH | 2 | 100 | L |
| 30 | M | 55 | BS | 2 | 12 | R |
| 31 | F | 54 | ALS | 1 | 120 | R |
| 32 | F | 66 | CA | 1 | 384 | L |
| 33 | F | 62 | ACS | 2 | 147 | L |
| 34 | M | 62 | RS | 2 | 0.5 | R |
| 35 | M | 58 | PSP | 1 | 60 | L |
| 36 | F | 37 | MC | 2 | 12 | R |
| 37 | M | 52 | RS | 2 | 3 | L |
| 38 | F | 63 | RS | 2 | 0.5 | L |
| 39 | M | 59 | TBI | 2 | 6 | L |
| 40 | F | 75 | RS | 2 | 4 | L |
| 41 | M | 53 | TBI | 2 | 11 | L |
| 42 | M | 70 | RS | 2 | 26 | L |
| 43 | F | 61 | CP | 2 | 732 | L |
| 44 | M | 79 | RS | 2 | 3 | L |
| 45 | M | 73 | RS | 2 | 8 | R |
| 46 | M | 74 | RS | 2 | 2 | L |
| 47 | F | 51 | LS | 2 | 23 | L |
| 48 | M | 66 | RS | 2 | 1.5 | L |
| 49 | M | 79 | RS | 2 | 3 | L |
| 50 | F | 69 | GBS | 2 | 4 | L |

Table 3. Data of included subjects. Diagnosis: MS= Multiple Sclerosis; LS: Left Stroke; RS= Right Stroke; BS= Bilateral Stroke; CA=Cerebellar Ataxia; ACS= Arnold-Chiari Syndrome; ALS= Amyotrophic Lateral Sclerosis; PD= Parkinson's Disease; SH=Subarachnoid Hemorrhage; TBI= Traumatic Brain Injury; MSA= Multiple System Atrophy; PSP= Progressive Supranuclear Palsy; MC= Meningo-cerebellitis; CP= Cerebral palsy; GBS= Guillan-Barré Syndrome. Dominance: L=Left; R=Right. Sex= M=male; F=female

Scorers

The SLT team of Hospital San Camillo volunteered to participate in this study as a scorer. The team, composed by 13 SLT, was divided into 2 groups according to their work experience. Group n°1 (“*skilled*”) was composed of 5 SLT who have worked with dysarthria more than 5 years; group n° 2 (“*beginners*”) consisted of 8 SLT whose work experience with dysarthria was lower than 5 years. Each participant was either on-line assessor or off-line scorer.

Procedure

The study consisted of 4 phases (displayed in figure 2):

- Phase 1 – *FOCUS GROUP AND TRAINING* (June – July 2015): the protocol was modified by means of focus group by the SLT team of Hospital San Camillo, following a literature review. Once the protocol was completed, the main researcher (i.e. De Biagi Francesca) administered the first assessment, which was video recorded. The SLT group was trained on the assessment and scoring methods by analyzing the video.

- Phase 2 – *SUBJECTS ENROLMENT* (August 2015 – May 2016): 50 subjects were recruited on the basis of the inclusion/exclusion criteria; assessments were video recorded. During this phase, data were stored by the main researcher (i.e. De Biagi Francesca) in an anonymized form; an alphanumeric code was attributed to each participant.

- Phase 3 – *PROTOCOL VALIDATION* (March – July 2016): Each video recording was scored twice from the main researcher (intra-rater reliability) and by one SLT of each group (inter-rater reliability). Accordingly, each SLT had to give scores for up to

10 subjects following protocol instructions and the training they had received. Patients' video recordings were assigned to scorers avoiding that the off-line scorer was not the same on-line scorer.

- Phase 4 – *STATISTICAL ANALYSIS* (July 2016): data were entered in an EXCEL spreadsheet and intra-rater and inter-rater agreement were evaluated by means of the Lin's coefficient along with the 95% confidence interval. Analysis was performed with SAS 9.4 (SAS Institute, Inc., Cary, NC, USA).

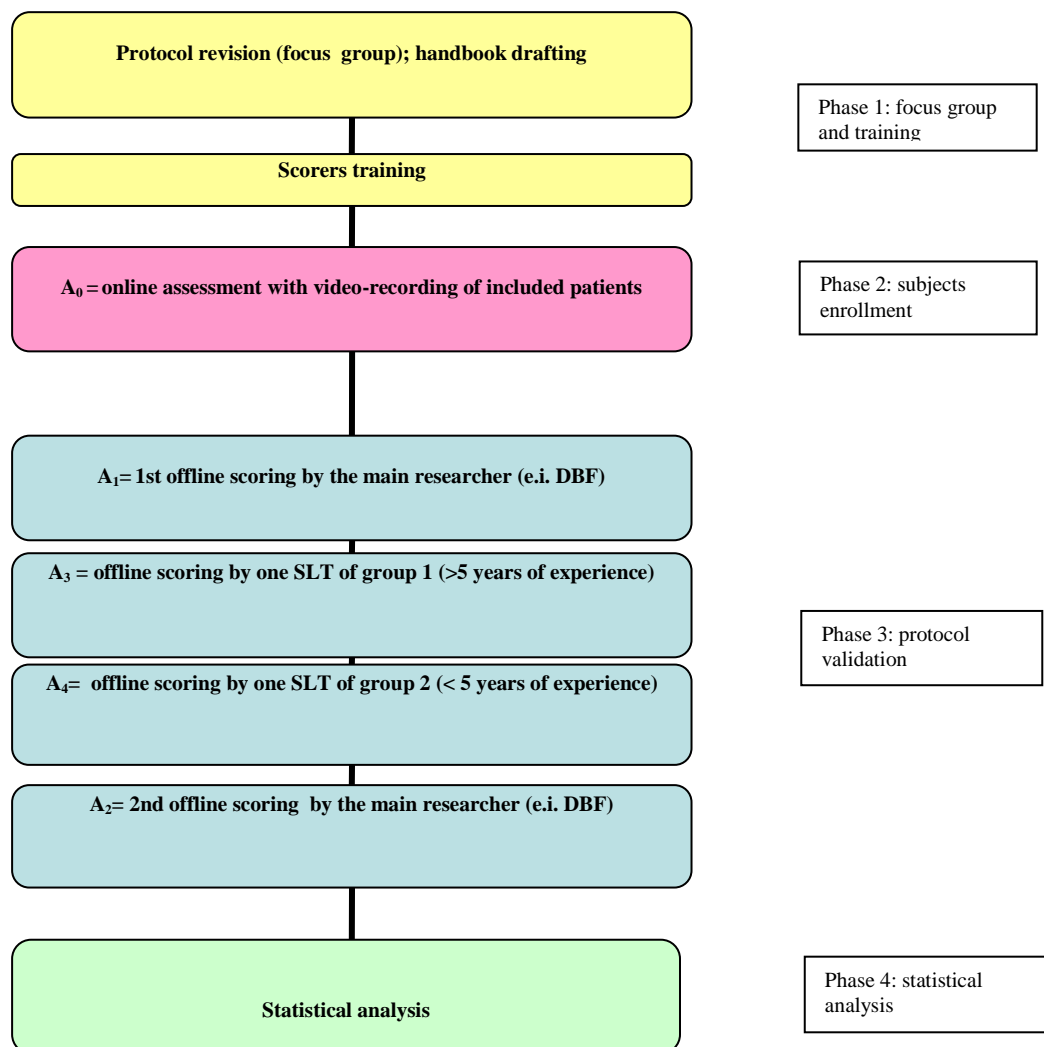


Fig 2. Study process flow-chart

Materials

The protocol (attached 1) is a modified short-form of “*Profilo di Valutazione della Disartria*” (Fussi, Cantagallo, Bertozzini, 1997) and is aimed to a perceptual analysis of the components that allow speech production. It is divided into 7 subscales, each composed by a different number of items: intelligibility, respiration, phonation, diadochokinesis, oral muscles, prosody and articulation. Differently from the original protocol, one subscale (“*reflexes*”) was completely removed and the total number of items was reduced from 71 to 35. This, according to the previous findings of the study of internal construct validity done by the same authors of the original scale (Cantagallo *et al.* 2006), in order to enhance the feasibility in a shorter time of administration. It was maintained the score system of the original protocol, that follow a 4-points Lickert scale (1= severe; 2= moderate; 3= mild; 4= normal) in order to use the same normative data (Cantagallo *et al.* 1997).

Following, a description of the scale:

➤ *Subscale A: intelligibility (2 items)*

Contextual intelligibility is assessed through a brief sample of spontaneous speech; signal-dependent intelligibility is evaluated through a brief excerpt of reading. Score is attributed following the categorization of the original protocol.

➤ *Subscale B: respiration (3 items)*

Two items evaluate expiratory (prolonged /s/) and phonatory (prolonged /a/) durations; Score is attributed following the normative data provided from the original protocol. One item assesses the degree of pneumonic-phonatory coordination. Qualitative information on respiration impairments is also annotated; but these data were not considered in the statistical analysis.

➤ *Subscale C: phonation (1 item):*

Patients are asked to self-assess the degree of fatigue while speaking following a 4-points lickert scale. The assessor should also take note of qualitative data of voice production (intensity, voice quality).

➤ *Subscale D: diadochokinesis (6 items)*

Patients are asked to repeat rapidly and accurately six different syllables; scores are attributed following normative data (number of syllables/5 seconds).

➤ *Subscale E: oral muscles (16 items)*

Muscular functionality of lips, tongue, jaw and soft palate is assessed in terms of motility, range of movements, rate and precision. Muscular strength was not taken into account for statistical analysis, due to feasibility problems.

➤ *Subscale F: prosody (4 items)*

Two items assess rhythm: patients are asked to repeat automatic series (months of the year) at a normal and a faster rate. Two items assess prosody: one item assesses the use of a normal intonation while speaking; another item assesses the ability of the patient to imitate different accents.

➤ *Subscale G: articulation (3 items)*

Two items assess the articulation and co-articulation of initial consonants and groups of consonants in the repetition of 44 words; one item assesses the repetition of the whole word (6 stimuli). Scores are given following normative data (number of correct words).

Results

Administration

All the attendee assessors and scorers felt confident with both the administration and the scoring method of the protocol. In fact, it was not necessary to implement any other retraining for them. Moreover, the time of the administration of the protocol was limited, ranging from a minimum of 8.43 minutes to a maximum of 30.1 minutes, with an average time of 17.07 minutes (SD=4.12 minutes). As expected, almost all the subjects included succeeded to complete the protocol in only one session. For only one patient, due to fatigue reasons, it was necessary to reschedule a second appointment to fulfill the assessment. Furthermore, all the included subjects, regardless of the severity and characteristics of the dysarthria, were able to perform almost all the subtest and the items of the protocol.

Offline scoring: intra-rater and inter-rater reliability

The offline intra-rater agreement was evaluated comparing, for each subject, two protocols scoring (A_1 and A_2) fulfilled by the main researcher of the study at two different times (t_1 and t_2). It was established a distance of 1 month between t_1 and t_2 in order to avoid the familiarity of the scorer with the assessments and the subjects.

The inter-rater agreement was estimated by analyzing the scores (A_1 , A_3 and A_4) from three different scorers, for each subject. A_1 was the score attributed from the main researcher; A_3 was provided from a scorer belonging to group 1 (SLT with more than 5 years of work experience) and A_4 was ascribed to a scorer belonging to group 2 (SLT with less than 5 years of work experience).

Table 4 resumes subjects' random allocation respectively to the online assessor (A_0) and offline scorers for intra-rater agreement ($A_1, A_2,$) and inter-rater agreement (A_1, A_3, A_4).

| SUBJECT | A0 | A1 | A2 | A3 | A4 |
|---------|----|----|----|----|----|
| 1 | 6 | 1 | 1 | 3 | 10 |
| 2 | 4 | 1 | 1 | 6 | 11 |
| 3 | 2 | 1 | 1 | 3 | 10 |
| 4 | 3 | 1 | 1 | 6 | 12 |
| 5 | 5 | 1 | 1 | 3 | 12 |
| 6 | 5 | 1 | 1 | 3 | 13 |
| 7 | 2 | 1 | 1 | 6 | 10 |
| 8 | 3 | 1 | 1 | 6 | 13 |
| 9 | 3 | 1 | 1 | 4 | 12 |
| 10 | 2 | 1 | 1 | 4 | 8 |
| 11 | 10 | 1 | 1 | 5 | 8 |
| 12 | 3 | 1 | 1 | 5 | 8 |
| 13 | 2 | 1 | 1 | 5 | 7 |
| 14 | 2 | 1 | 1 | 5 | 7 |
| 15 | 2 | 1 | 1 | 5 | 10 |
| 16 | 3 | 1 | 1 | 5 | 10 |
| 17 | 3 | 1 | 1 | 6 | 7 |
| 18 | 6 | 1 | 1 | 2 | 7 |
| 19 | 5 | 1 | 1 | 4 | 12 |
| 20 | 3 | 1 | 1 | 4 | 12 |
| 21 | 5 | 1 | 1 | 6 | 12 |
| 22 | 5 | 1 | 1 | 6 | 12 |
| 23 | 2 | 1 | 1 | 4 | 12 |
| 24 | 10 | 1 | 1 | 2 | 12 |
| 25 | 10 | 1 | 1 | 2 | 13 |
| 26 | 4 | 1 | 1 | 2 | 10 |
| 27 | 4 | 1 | 1 | 6 | 13 |
| 28 | 2 | 1 | 1 | 3 | 10 |
| 29 | 10 | 1 | 1 | 2 | 13 |
| 30 | 10 | 1 | 1 | 3 | 9 |
| 31 | 2 | 1 | 1 | 3 | 10 |
| 32 | 2 | 1 | 1 | 3 | 10 |
| 33 | 4 | 1 | 1 | 3 | 10 |
| 34 | 6 | 1 | 1 | 3 | 10 |
| 35 | 3 | 1 | 1 | 2 | 9 |
| 36 | 5 | 1 | 1 | 2 | 9 |
| 37 | 3 | 1 | 1 | 2 | 9 |
| 38 | 4 | 1 | 1 | 6 | 9 |
| 39 | 5 | 1 | 1 | 6 | 8 |
| 40 | 6 | 1 | 1 | 5 | 8 |
| 41 | 3 | 1 | 1 | 5 | 7 |
| 42 | 2 | 1 | 1 | 5 | 7 |
| 43 | 10 | 1 | 1 | 5 | 7 |
| 44 | 5 | 1 | 1 | 2 | 7 |
| 45 | 6 | 1 | 1 | 2 | 8 |
| 46 | 6 | 1 | 1 | 4 | 8 |
| 47 | 6 | 1 | 1 | 4 | 13 |
| 48 | 2 | 1 | 1 | 4 | 13 |
| 49 | 6 | 1 | 1 | 5 | 13 |
| 50 | 13 | 1 | 1 | 5 | 8 |

Table 4. Random table for assessors (A_0) and offline scorers (A_1, A_2, A_3, A_4) allocation. Main researcher: 1= DBF. Scorers of group 1: 2= SN, 3=JF, 4=VR, 5=GB; 6=SN. Scorers of group 2: 7=IK, 8=MG, 9=IB, 10=SB, 11=AF, 12=FB, 13=AC

The Concordance Correlation Coefficient (CCC) was estimated by means of the Lin's coefficient with a 95% Confidence Interval (95%CI). Analyses were performed taking into account the total

scores of the seven subscales of the protocol, moreover items 1 and 2 (respectively “*Contextual intelligibility*” and “*Signal-dependent intelligibility*”) were also considered independently due to their clinical specificity as functional outcome measure for dysarthria severity. Missing data were not imputed because their prevalence did not affect the sample size significantly.

The offline intra-rater and inter-rater agreement results are reported and compared in table 5.

| SUBSCALE | | Offline Intra-rater agreement (A ₁ - A ₂) | | Offline Inter-rater agreement (A ₁ - A ₃ - A ₄) | |
|----------|------------------------|--|--------------------|---|--------------------|
| | | N | CCC (95%CI) | N | CCC (95%CI) |
| A | Intelligibility | 49 | 0.89 (0.80 - 0.95) | 49 | 0.63 (0.48 - 0.74) |
| | - Signal-dependent | 49 | 0.85 (0.71 - 0.94) | 49 | 0.59 (0.43 - 0.72) |
| | - Contextual | 50 | 0.81 (0.70 - 0.92) | 50 | 0.57 (0.43 - 0.67) |
| B | Respiration | 49 | 0.90 (0.85 - 0.94) | 47 | 0.88 (0.83 - 0.93) |
| C | Phonation | 45 | 1 | 42 | 0.89 (0.76 - 0.96) |
| D | Diadochokinesis | 46 | 0.96 (0.90 - 0.98) | 45 | 0.81 (0.73 - 0.87) |
| E | Oral muscles | 46 | 0.87 (0.76 - 0.93) | 42 | 0.75 (0.60 - 0.87) |
| F | Prosody | 49 | 0.84 (0.69 - 0.92) | 48 | 0.72 (0.63 - 0.86) |
| G | Articulation | 49 | 0.94 (0.88 - 0.97) | 49 | 0.74 (0.63 - 0.84) |

Table 5. Offline intra-rater and inter-rater agreement; CCC = Concordance Correlation Coefficient; 95%CI = 95% Confidence Interval;

As speculated, for each subscale the intra-rater agreement was satisfactory and confirmed the alternative hypothesis (Fig. 3). As a matter of fact, all the subareas had a CCC higher than 0.8 with a narrow CI. Almost all the CI upper limits were above 0.9 and for only one subscale (“*Prosody*”) the lower limit was inferior than 0.7. Obviously, the subscale C “*Phonation*” has a perfect concordance because it was a self-reported measure; however this

measure seemed to be not stable when assessed by different scorers.

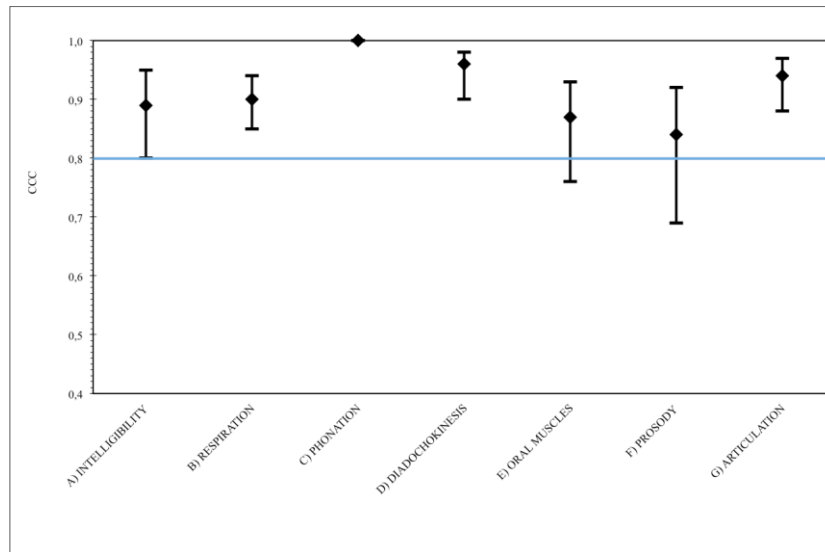


Figure 3. Offline intra-rater agreement between A_1 - A_2 ; CCC (95%CI)

Contrary to our expectations, there was a high-grade inter-rater agreement only for 3 subscales: B “Respiration”, C ”Phonation” and D ”Diadochokinesis” (Fig. 4). Furthermore, for each subscale the CI was wider and for one subscale (A “Intelligibility”) the CI lower limit was even lower than 0.5. These results were partially attributed to the presence of more missing data than in the intra-rater analysis; in fact, for some subscale, only 42 subjects were analyzed.

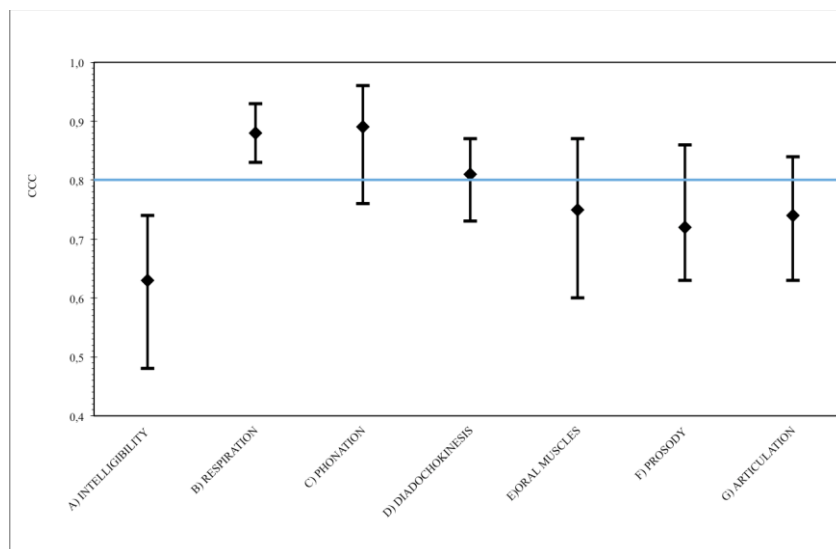


Figure 4. Inter-rater agreement between A_1 - A_3 - A_4 ; CCC (95%CI)

The inter-rater agreement was also analyzed in relation to the expertise and acquaintance with the protocol. The CCC was estimated between the scores given by the main researcher (A₁) and a “*Skilled*” SLT (A₃) and between a “*Beginner*” SLT (A₄). These analyses were performed in order to understand if the knowledge of the protocol could have affected the ability to rate. Table 6 displays the results.

| SUBSCALE | | Offline Inter-rater agreement (A ₁ – A ₃) | | Offline Inter-rater agreement (A ₁ - A ₄) | |
|----------|------------------------|--|--------------------|--|--------------------|
| | | N | CCC (CI 95%) | N | CCC (CI 95%) |
| A | Intelligibility | 49 | 0.69 (0.54 - 0.78) | 49 | 0.67 (0.50 - 0.80) |
| | - Signal-dependent | 49 | 0.68 (0.53 - 0.80) | 49 | 0.57 (0.34 - 0.74) |
| | - Contextual | 50 | 0.61 (0.48 - 0.74) | 50 | 0.65 (0.48 - 0.79) |
| B | Respiration | 47 | 0.89 (0.84 - 0.94) | 48 | 0.87 (0.80 - 0.92) |
| C | Phonation | 43 | 0.90 (0.65 - 0.98) | 43 | 0.87 (0.73 - 0.94) |
| D | Diadochokinesis | 45 | 0.84 (0.75 - 0.91) | 46 | 0.73 (0.61 - 0.83) |
| E | Oral muscles | 43 | 0.75 (0.55 - 0.87) | 44 | 0.77 (0.60 - 0.89) |
| F | Prosody | 48 | 0.71 (0.58 - 0.85) | 49 | 0.73 (0.60 - 0.84) |
| G | Articulation | 49 | 0.76 (0.65 - 0.86) | 49 | 0.67 (0.52 - 0.80) |

Table 6. Offline inter-rater agreement, difference between “skilled” and “beginners” scorers; CCC = Concordance Correlation Coefficient; 95%CI = 95% Confidence Interval;

As it is shown in table 6, there was a high-grade CCC between the main researcher and a “*beginner*” SLT in only 2 subscales of the protocol (“*Respiration*” and “*Phonation*”). The same results were found also between a “*skilled*” SLT and the main researcher, where the agreement was found to be satisfactory for only 2 subscales (“*Respiration*” and “*Phonation*”). Figure 5 and figure 6 show that the level of agreement was not worse than 0.5 for all

the items, and for “*Respiration*” it was even equal or better than 0.8.

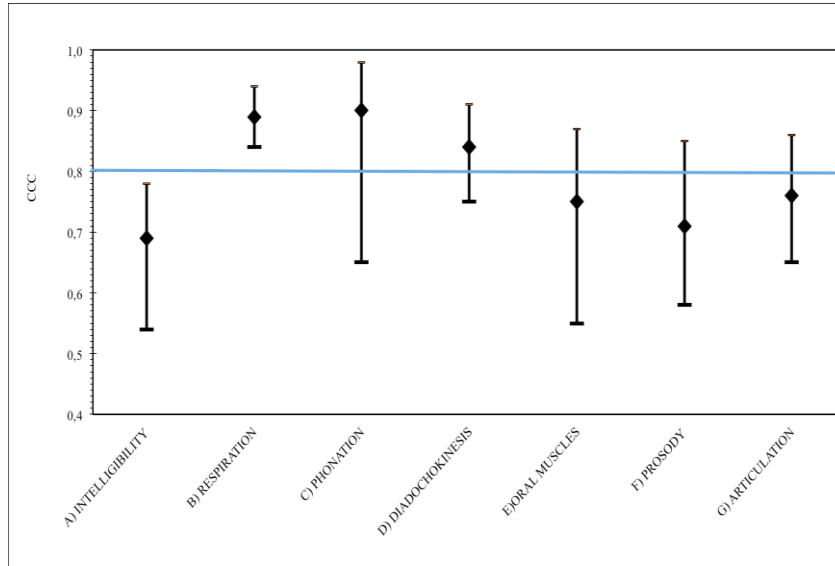


Figure 5. Inter-rater agreement between A_1 - A_3 ; CCC (IC 95%)

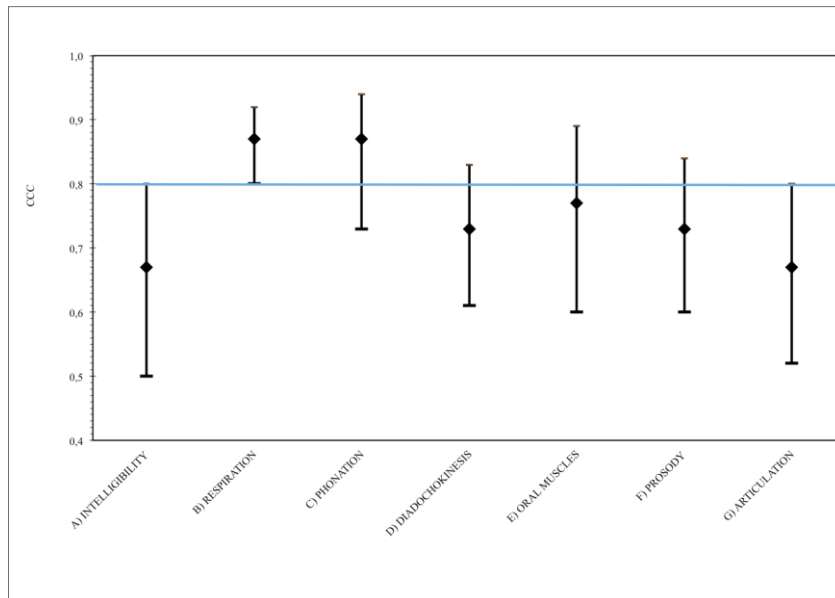


Figure 6. Inter-rater agreement between A_1 - A_4 ; CCC (IC 95%)

Online scoring: inter-rater reliability

The intra and inter-rater reliability were assessed analyzing the data coming from offline scoring, obtained watching subjects' video recording. Despite the protocol is a *face-to-face* assessment the procedure based on video recording was designed with the aim to replicate a feasible setting within the SLT service at Hospital San Camillo. Thus, it was analyzed the online inter-rater reliability estimating the CCC between online assessment (A_0) and one of the measure of the offline scorers (A_1). It was chosen A_1 because these data were produced by the main researcher of the study, who, in this case was considered as the “gold standard” for the assessment. Table 7 exhibits the results.

| Online/offline inter-rater agreement (A_0- A_1) | | |
|--|----------|---------------------|
| SUBSCALE | N | CCC (CI 95%) |
| Intelligibility | 49 | 0.76 (0.63 - 0.86) |
| - Signal-dependent | 49 | 0.69 (0.59 - 0.81) |
| - Contextual | 50 | 0.73 (0.51 - 0.86) |
| Respiration | 49 | 0.76 (0.61 - 0.87) |
| Phonation | 43 | 0.75 (0.45 - 0.90) |
| Diadochokinesis | 46 | 0.66 (0.42 - 0.81) |
| Oral muscles | 46 | 0.67 (0.42 - 0.83) |
| Prosody | 49 | 0.69 (0.53 - 0.79) |
| Articulation | 49 | 0.62 (0.45 - 0.78) |

Table 7. Online/offline inter-rater agreement; CCC = Concordance Correlation Coefficient; 95%CI = 95% Confidence Interval;

As it is presented in figure 7, any subscale of the protocol showed an excellent agreement between the two scoring modalities (online/offline). In fact, the CI were wider and, although all the CI upper limit were above 0.7, the lower limits could be even below 0.5.

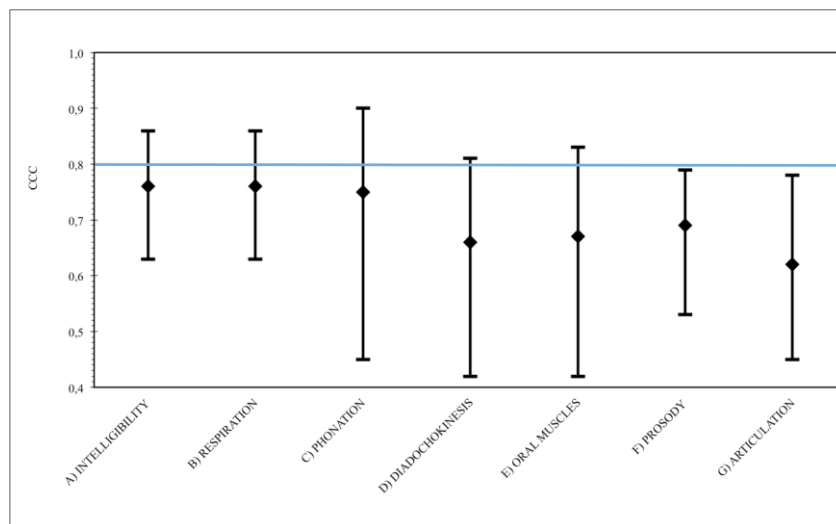


Figure 7. Inter-rater agreement between A0-A1; CCC (IC 95%)

Discussion

Psychometric features

The study aimed to explore the reliability of the protocol, analyzing the consistency in measurements between the same rater and among different raters with different level of expertise. The intra-rater concordance was found to be very high, with a CCC more than 0.8 for many subscales and a tight CI. One subscale (“*Phonation*”), which was the only self-reported measure, even performed a perfect concordance. Besides statistical findings, qualitatively, there were no significant differences among the performances of the different subscales. These data entail that the scoring system was stable in measurements repeated over time. Basing solely on these data, it

could be concluded that the protocol is a reliable tool to track patient's progress to determine the effectiveness of the treatment. However, the inter-rater agreement was found to be worse than the intra-rater one. In fact, only three subscales of the protocol had a high CCC. Not surprisingly, two of these subscales ("*Respiration*" and "*Diadochokinesis*") are the most objective measurements, with clear normative data to give scores. Whereas, one of these three subscales is a self-reported measure. Nevertheless, beyond statistical significance, in a qualitative perspective, the agreement for the other subscales could be considered satisfactory as well. As a whole, the CCC for the all other subscale ranges from 0.63 to 0.75, with almost all the CI lower limits above 0.6 and all the upper limits all above 0.8. This except from "*Intelligibility*", and its two items analyzed, which was the subscale that had the worst agreement among all, with a CI lower limit minor than 0.5. These results are not favorable if considered the importance of this variable as a functional outcome.

At any rate, these data indicate that four subscales on seven are susceptible to subjective judgment. Thus, it is possible that the scoring system or the measurement of some items should be revised in order to make it as more objective as possible.

Other analyses were performed in order to understand whether the discrepancies between inter and intra-rater reliability could have been related to the expertise of the raters. However, no significant difference was found comparing expert to non-skilled raters, thus inconsistency might be attribute other reasons than lack of training or knowledge of the instrument by final users.

Moreover, it was found a poor inter-rater agreement between online and offline scoring. This finding may arise questions about the suitability of administering this tool by video recording, then face-to-face for assessment. However, once again, qualitative analyses showed moderate concordance.

Clinical utility

The results revealed a worthy clinical utility of the protocol.

First of all, the protocol provides a complete assessment of the meaningful parameters for the assessment of motor speech disorders. The instrument provides indeed a measurement of the functionality of speech structures, a perceptual analysis of speech and judgment of intelligibility.

The time of administration of the protocol is indeed limited, proving to be suitable for the clinical practice expected timing. Moreover, the time of administration was also sufficient enough in relation to patients' abilities. In fact, almost all the included subjects managed to complete the assessment in only one session. These results are quite satisfactory and confirmed the need to use a short-form of the original protocol, as speculated by the authors themselves.

Moreover, the protocol, that has a limited cost, does not require any technical or specific equipment. The only resource that should be accounted for the administration is the time of the healthcare professional, which is strongly recommended to be a trained SLT.

Besides, the protocol seemed easy to administer, as all the assessors and the scorers did not solicit for a retraining. Although a specific training seems to be unavoidable, it can be settled in a couple of meetings of few hours. These two features also imply limited organizational constraints.

Finally, the difficulty of the protocol's items seemed to be adequate for both the different kinds of dysarthria and the various severity levels. As a matter of fact, almost all the included subjects were able to complete almost all the items.

A limit to the clinical utility is that the protocol is a clinician-rated performance instrument. On one hand, a clinician-rated tool should presuppose a qualitatively and quantitatively richer

measure, conceptually based on a solid theoretic construct. On the other hand, this type of measure implies more burdens for the professionals, as well as more potential raters' biases and errors. Moreover the patients' perspective is not considered and this could be a potential bias while settling the goals of the treatment. Although it should be considered that patients' perception of their impairment could be often unreliable, due to cognitive disorders, such as awareness deficits, that frequently occur in neurologic patients.

Limitations

As declared, the intent was to proceed with an exploratory pilot study. Thus, the sample size was limited to 50 subjects in order to allow the feasibility for SLT service along the pre-determined duration of the study (1 year). This implies that the generalizability of the findings of this study is limited by its small sample size as well as its poor stratification among the different diagnoses that may result in dysarthria. Moreover, both subjects and raters were enrolled in the same center. In fact, even the common background of the assessors and the raters could have been a bias in the validation process.

As a matter of fact, one of the purposes of the study was to find preliminary results that could be used to estimate the adequate sample size for future studies aimed to develop the Italian validation of the protocol.

Increasing sample size and diversity in the types of clients examined, together with the recruitment of raters and assessors from other centers, might be useful in minimizing the possible biases.

Conclusions

The aim of the study was to investigate the intra and inter-rater reliability of a short-form protocol to assess dysarthria in neurologic patients. Although further analyses are required, our preliminary data showed an excellent consistency of ratings in repeated measures over time. Nevertheless, it was highlighted a scarce stability of ratings among different scorers, especially in the online administration of the protocol. As a matter of fact, many items of the protocol were found to be susceptible to individual's judgment. Above all, the measurement of the functional outcome (i.e. intelligibility) seemed to be unsatisfactory. The discrepancy between inter and intra-rater reliability was not attributed to the level of acquaintance with the protocol. Thus, it may be postulated that the scoring system itself, as well as the normative data, should be reconsidered.

The study revealed an adequate clinical utility of the protocol, whose administration has been thought to be convenient and affordable in terms of duration and resources required. Besides, the protocol with regard to parameters assessed and items' difficulty seemed to be adequate for the different type and level of dysarthria severity. However, it should be taken into consideration the limitations of the protocol as a clinician-related measure. In fact, the client's perspective and expectations are not considered. Moreover, the protocol is aimed to assess speech impairment, while the levels of disability and participation are not considered. This may be in contrast with the latest research that emphasizes participation-focused assessments and interventions. In conclusion, while the protocol appears to be a potentially useful test, the study warrants cautious interpretation, due to the limited generalizations of the findings. Further research is indeed required in order to validate the instrument, possibly integrating it with other types of outcome measures.

Future studies are needed to foster the use of standardized and validated tool to assess outcomes in rehabilitation. A grounded measurement of the outcomes is undeniably important in order to establish the patient's baseline status and monitor his improvements, determining the usefulness of treatments.

In this way, outcome measures reporting may contribute to improve clinical practice, supporting organizational changes and leading to efficient policy acts.

References

1. Bloch, S & Wilkinson, R. (2011) Acquired dysarthria in conversation: methods of resolving understandability problems. *Int J Lang Commun Disord.* 46(5):510-523
2. Cancialosi, P, Basagni, B. (2011) Disturbi del linguaggio e della comunicazione. In: Mazzucchi A. *La riabilitazione delle gravi cerebrolesioni acquisite*. Percorsi sanitario-assistenziali, complessità gestionale, evidenza dei risultati. Ed. Giunti OS. Firenze
3. Cantagallo, A, La Porta, F, Abenante, L. (2006). Dysarthria assessment; Robertson profile and self-assessment questionnaire. *Acta Phon Lat.* 28:246-61.
4. Darley, FL, Aronson, AE, Brown, JR. (1969). Clusters of deviant speech dimensions in the dysarthrias. *J speech Hear Res.* 12:462-496
5. Darley, FL, Aronson, AE, Brown, JR. (1969). Differential diagnostic patterns of dysarthrias. *J Speech Lang Hear Res.* 12:246-269
6. Dos Santos Barreto, S, Ortiz, KZ. (2008). Intelligibility measurements in speech disorders: a critical review of the literature. *Pro-Fono Revista de Atualização Científica.* 20(3):201-6
7. Duffy, JR. (2013). *Motor Speech Disorders*. Substrates, Differential Diagnosis and Managment. Mosby. St. Louis
8. Enderby, P. (2013). Disorders of communication: dysarthria. In Barnes, MC, Good, DC.. *Neurological Rehabilitation*. Elsevier B.V. Amsterdam
9. Flowers, HL, Silver, FL, Fang, J, Rochon, E, Martino, R (2013). The incidence, co-occurrence, and predictors of dysphagia, dysarthria, and aphasia after first-ever acute ischemic stroke. *J Commun Disord.* 46(3):238-48
10. Fussi, F, Cantagallo, A. (1997). *Profilo di valutazione della disartria*. Omega. Torino

11. Hartelius, L, Runmarker, B, Andersen, O. (2000) Prevalence and characteristics of dysarthria in a multiple-sclerosis incidence cohort: relation to neurological data. *Folia Phoniatr Logop.* 52(4):160-77
12. Hartelius, L, Svensson, P. (1994). Speech and swallowing symptoms associated with Parkinson's disease and multiple sclerosis: a survey. *Folia Phoniatr Logop.* 46:9–17.
13. Hegde, MN, Freed, D. (2011). Assessment of Dysarthria. In: Hegde, MN, Freed, D. *Assessment of Communication Disorders in Adults*. Plural Publishing. San Diego.
14. Hill, AJ, Theodoros, DG, Russell, TG & Ward, EC. (2009). The Redesign and Re-evaluation of an Internet-Based Telerehabilitation System for the Assessment of Dysarthria in Adults. *Telemedicine and e-Health.* 15(9):840-850
15. Kent, RD, Vorperian, HK, Kent, JF, Duffy, JR. (2003). Voice dysfunction in dysarthria: application of the Multi-Dimensional Voice Program. *Journal of Communication Disorders.* 36:281-306
16. Kent, RD; Volperian, HK, Duffy, JR. (2003). Voice dysfunction in dysarthria: application of the Multi-Dimensional Voice Program™. *Journal of Communication Disorders.* 36(4):281-306
17. Kim, J, Kent, RD, Weismerv, G. (2011). An acoustic study of the relationships among neurologic disease, dysarthria type and severity of dysarthria. *J Speech Lang Hear Res.* 54:417-429
18. Lin, L, Hedayat, AS, Sinha, B & Yang M. (2002). Statistical Methods in Assessing Agreement. *JASA.* 97(457):257-270.
19. Lin, L, Hedayat, AS, Wu, W. (2012). *Statistical Tools for Measuring Agreement*. Springer. New York
20. Lin, L, I-Kuei, L. (1989). A Concordance Correlation Coefficient to Evaluate Reproducibility. *Biometrics.* 45:255-268.
21. Lin, L, I-Kuei, L. (1992). Assay Validation Using the Concordance Correlation Coefficient. *Biometrics.* 48:599-604.

22. Lin, L, I-Kuei, L. (2000). A Note on the Concordance Correlation Coefficient. *Biometrics*. 56:324-325.
23. Mackenzie, C, Muir, M, Allen, C & Jensen, A. (2014). Non-speech oro-motor exercises in post-stroke dysarthria intervention: a randomized feasibility trial. *Int J Lang Commun Disord*. 49(5):602-617
24. Miller, N (2013). Measuring up to speech intelligibility. *Int J Lang Commun Disord*. 48(6):601-612
25. Miller, N, Allcock, L, Jones, D, Noble, E, Hildreth, AJ, & Burn, DJ. (2007). Prevalence and pattern of perceived intelligibility changes in Parkinson's disease. *Neurol Neurosurg Psychiatry*. 78(11):1188–119
26. Nicastri, M, Chiarella, G, Gallo, LV, Catalano, M, Cassandro, E. (2004). Multidimensional Voice Program (MDVP) and amplitude variation parameters in euphonic adult subjects. Normative study. *Acta Otorhinolaryngol Ital*. 24(6):337-41
27. Patel, R, Usher, N, Kember, H, Russel, S, Laures-Gore, J. (2014). The influence of speaker and listener variables on intelligibility of dysarthric speech. *Journal of Communication Disorders*. 51:13-18
28. Pezzella, FR, Anticoli, S, Sorrentino, R. (2013). Fisiopatologia dei disturbi motori della parola. In: Ruoppolo, G, Amitrano, A. a cura di. *Disartria: possiamo fare di più? Relazione Ufficiale Congresso nazionale della Società Italiana di Foniatria e Logopedia*. Omega Edizioni. Torino
29. Piacentini, V, Mauri, I, Cattaneo, D, Gilardone, M, Montesano, A, Schindler, A. (2014). Relationship Between Quality of Life and Dysarthria in Patients With Multiple Sclerosis. *Archives of Physical Medicine and Rehabilitation*. 95:2047-54
30. Pinto, S, Ozsancak, C, Tripoliti, E, Thobois, S, Limousin-Dowsey, P & Auzou, P. (2004). Treatments for dysarthria in Parkinson's Disease. *The Lancet Neurology*. 3:547-556

31. Robertson, SJ. (1982). *Dysarthria profile*. Communication Skill Builders. Tucson
32. Scettino, I, Albino, F, Ruoppolo, G. (2013). La valutazione bedside del soggetto disartrico. In: Ruoppolo, G, Amitrano, A. a cura di. *Disartria: possiamo fare di più? Relazione Ufficiale Congresso nazionale della Società Italiana di Foniatria e Logopedia*. Omega Edizioni. Torino
33. Schalling, E, Hartelius, L. (2013). Speech in spinocerebellar ataxia. *Brain & Language*. 127:317-322
34. Sellars, C, Hughes, t, Langhorne, P. (2005). Speech and language therapy for dysarthria due to non-progressive brain damage. *Cochrane Database of Systematic Reviews*. Published by John Wiley & Sons, Ltd.
35. Solomon, NP, Clark, HM, Makashay, MJ, Newman, LA. (2008). Assessment of Orofacial Strength in Patients with Dysarthria. *J Med Speech Lang Pathol*. 16(4):251–258
36. Urban, PP. (2013). Speech motor deficits in cerebellar infarctions. *Brain & Language*. 127:323-326
37. Walshe, M, Miller, N, Leahy, M, Murray, A. (2008). *Intelligibility of dysarthric speech: perceptions of speakers and listeners*. International Journal of Language & Communication Disorders. 43(6):633-648
38. Wang, YT, Kent, RD, Duffy, JR, Thomas, JE. (2005). Dysarthria associated with traumatic brain injury: speaking rate and emphatic stress. *J Commun Disord*. 38(3):231-60.
39. Wang, EQ. (2010). Dysarthria. In Kompoliti, K, Metman, LV. *Encyclopedia of Movement Disorders*, Volume 1. Elsevier LTD. San Diego, CA
40. Wannberg, P, Schalling, E, Hartelius, L. (2015). Perceptual assessment of dysarthria: Comparison of a general and a detailed assessment protocol. *Logoped Phoniatr Vocol*. 7:1-9

41. Yorkston, KM. (2007). The Degenerative Dysarthrias: A Window into Critical Clinical and Research Issues. *Folia Phoniatr Logop.* 59:107–117

Acknowledgments

This thesis benefited from the help, support and guidance of many persons. First of all, I would like to thank the main supervisor, Professor Anna Chiara Frigo, statistician at Department of Cardiac, Thoracic and Vascular Sciences of University of Padova. Her professionalism as well as her methodological competence was fundamental to the birth and the development of the project.

Secondly, I would express my gratitude for the continue support to my colleague, Andrea Turolla, physiotherapist coordinator of the Laboratory of Kinematics and Robotics of Hospital San Camillo.

Moreover, I would like to thank Dr. Francesca Meneghello, head of the Neuropsychology Department of Hospital San Camillo, for promoting the project.

Furthermore, a heartfelt thanks to all the other collaborators, the SLT team at Hospital San Camillo. Giulia, Sara, Virginia, Sara, Martina, Irene, Isabella, Jessica, Silvia, Federica, Alessandra and Agnese are marvellous colleagues. My appreciation is going to them not only for the great work they have done on this research, but mostly for being the driving force in my professional growth.

Moreover, my gratefulness to all my friends and to Francesco, for always being there in these two tough years of study and work.

At least but not last, my gratitude to my family, mum, dad and Marleen, for the economic support, but mostly for having lighted in me the spark of curiosity and for having always fostered a culture of learning and intellectual freedom.

Enclosed

Protocollo di Valutazione Disartria e Disfonia

Vers. Mod. da A. Cantagallo e F. Fussi (1997)

riadattata dal Servizio di Neuropsicologia dell'I.R.C.C.S. S. Camillo di Venezia (2015)

Nome e Cognome _____ Data _____

Diagnosi _____ Esame n° _____

Esordio _____ Operatore _____

Prevalenza emisferica _____

Note _____

| AREE DI VALUTAZIONE | | |
|---------------------|--------------------------|--------|
| A | Intelligibilità | ___/8 |
| B | Respirazione | ___/12 |
| C | Fonazione | ___/4 |
| D | Diadococinesi | ___/24 |
| E | Muscolatura oro-facciale | ___/64 |
| F | Prosodia | ___/16 |
| G | Articolazione | ___/12 |

A. Intelligibilità dell'eloquio

| | | | |
|---|--|--|---------------------------------|
| 1 | 2 | 3 | 4 |
| 1 | 2 | 3 | 4 |
| l'eloquio è spesso inintelligibile, ma con una ripetizione più accurata può essere compreso | generalmente intelligibile, ma l'ascoltatore deve fare molta attenzione. Alcune parole hanno bisogno di essere ripetute e, se la stanza è rumorosa o l'argomento sconosciuto, c'è difficoltà di comprensione | leggera difficoltà ma sempre intelligibile | nessuna alterazione osservabile |

→ **Letture brano "Notturmo" – allegato n°2 e breve conversazione "Mi racconti come si fa il caffè"**

| B. Respirazione | | | | |
|---------------------------------------|-----------|-------------------|-------------|---------------------|
| | Sternale | Costale Superiore | Mista | Costo Diaframmatica |
| Riposo | | | | |
| Conversazione | | | | |
| Voce Proiettata | | | | |
| Durata Espiratoria /s:/ | 1 (1"-9") | 2 (10"-14") | 3 (15"-19") | 4 (20"-30") |
| Durata Fonatoria /a:/ | 1 (1"-5") | 2 (6"-10") | 3 (11"-14") | 4 (15"-25") |
| Incoordinazione pneumofonica in conv. | 1 (grave) | 2 (media) | 3 (lieve) | 4 (assente) |

→ **ripetere l'esecuzione 2 volte e segnare la prestazione migliore**

| C. Fonazione (in conversazione) | | | | |
|---------------------------------|---------------------------------|-----------------------------------|----------------------------------|--------------------------------|
| Attacco Vocale | Duro <input type="checkbox"/> | Dolce <input type="checkbox"/> | Afono <input type="checkbox"/> | Altro <input type="checkbox"/> |
| Qualità Vocale | * | | | |
| Intensità | Debole <input type="checkbox"/> | Adeguata <input type="checkbox"/> | Elevata <input type="checkbox"/> | Altro <input type="checkbox"/> |
| Affaticabilità riferita | 1 (molto) | 2 (abbastanza) | 3 (poco) | 4 (per niente) |

* normale, rauca, soffiata, debole, pressata, diffonica, con break vocali, stridente, nasale, ingolata, in registro di falsetto, piena, con vocal fry, sfinterica, variabile...

→ **"ritiene che la sua voce si affatichi durante il giorno?"**

D. Diadococinesi

| | | | | |
|-------------------------------|---------|----------|-----------|-----------|
| Ripetere "u-i" rapidamente | 1 (1-4) | 2 (5-9) | 3 (10-14) | 4 (15-20) |
| Ripetere "pa" rapidamente | 1 (1-7) | 2 (8-14) | 3 (15-19) | 4 (20-30) |
| Ripetere "ta" rapidamente | 1 (1-7) | 2 (8-14) | 3 (15-19) | 4 (20-30) |
| Ripetere "ka" rapidamente | 1 (1-7) | 2 (8-14) | 3 (15-19) | 4 (20-30) |
| Ripetere "kala" rapidamente | 1 (1-4) | 2 (5-9) | 3 (10-14) | 4 (15-20) |
| Ripetere "p.t.k." rapidamente | 1 (1-3) | 2 (4-7) | 3 (8-11) | 4 (12-15) |

→ **Segnare il numero di ripetizioni in 5"**

E. Muscolatura Facciale

| | | | | | |
|--|--|---------------------------------------|--|---|---|
| Labbra | Aspetto | Rilassate <input type="checkbox"/> | Tese <input type="checkbox"/> | Atrofiche <input type="checkbox"/> | |
| | | Deviate a dx <input type="checkbox"/> | | Deviate a sx <input type="checkbox"/> | |
| | Forza * | 1 | 2 | 3 | 4 |
| | Velocità di chiusura | 1 | 2 | 3 | 4 |
| | Estensione | 1 | 2 | 3 | 4 |
| | Protrusione | 1 | 2 | 3 | 4 |
| | Tono | Normotono <input type="checkbox"/> | Ipotono <input type="checkbox"/> | Iperono <input type="checkbox"/> | |
| Scialorrea | Sì <input type="checkbox"/> | | No <input type="checkbox"/> | | |
| Lingua | Aspetto | Normale <input type="checkbox"/> | Atrofica <input type="checkbox"/> | Fascicolazioni <input type="checkbox"/> | |
| | | Deviate a dx <input type="checkbox"/> | Deviate a sx <input type="checkbox"/> | Rigida <input type="checkbox"/> | |
| | Forza contro resistenza * | 1 | 2 | 3 | 4 |
| | Motilità | 1 | 2 | 3 | 4 |
| | Velocità di movimento | 1 | 2 | 3 | 4 |
| | Protrusione | 1 | 2 | 3 | 4 |
| | Arretramento (/k/; /g/) | 1 | 2 | 3 | 4 |
| | Lateralizzazione sx | 1 | 2 | 3 | 4 |
| | Lateralizzazione dx | 1 | 2 | 3 | 4 |
| | Innalzamento (/l/) | 1 | 2 | 3 | 4 |
| | Capacità di seguire l'arcata superiore | 1 | 2 | 3 | 4 |
| Capacità di seguire l'arcata inferiore | 1 | 2 | 3 | 4 | |
| Velo | Elevazione e retrazione | Simmetrico <input type="checkbox"/> | | Asimmetrico <input type="checkbox"/> | |
| | Tenuta d'elevazione | Normale <input type="checkbox"/> | Fuga d'aria <input type="checkbox"/> | Rino chiusa <input type="checkbox"/> | |
| Mandibola | Aspetto | Normale <input type="checkbox"/> | Deviazione dx <input type="checkbox"/> | Deviazione sx <input type="checkbox"/> | |
| | Tono | Normotono <input type="checkbox"/> | Cadente <input type="checkbox"/> | Serrata <input type="checkbox"/> | |
| | Apertura/chiusura | 1 | 2 | 3 | 4 |
| | Lateralizzazione a dx | 1 | 2 | 3 | 4 |
| | Lateralizzazione a sx | 1 | 2 | 3 | 4 |
| | Protrusione | 1 | 2 | 3 | 4 |
| | Forza contro resistenza * | 1 | 2 | 3 | 4 |

* non conteggiato negli indicatori riassuntivi

F. Prosodia

| | | | | |
|---|---|---|---|---|
| Mantenimento del ritmo adeguato | 1 | 2 | 3 | 4 |
| Aumento della velocità | 1 | 2 | 3 | 4 |
| Uso dell'intonazione adeguata | 1 | 2 | 3 | 4 |
| Imitazione di differenti modalità di accentazione | 1 | 2 | 3 | 4 |

→ **Chiedere i mesi dell'anno**

→ **Chiedere i mesi dell'anno più velocemente possibile**

→ **In conversazione**

→ **GIOVANNI è venuto domenica**

→ **capitàno – càpitano - capitanò**

Giovanni E' VENUTO domenica

→ **no? Noooo! NO**

Giovanni è venuto DOMENICA

G. Articolazione

| | | | | |
|------------------------------------|----------|-----------|-----------|-----------|
| Ripetizione di consonanti iniziali | 1 (1-11) | 2 (12-17) | 3 (18-20) | 4 (21-22) |
| Ripetizione di gruppi consonantici | 1 (1-11) | 2 (12-17) | 3 (18-20) | 4 (21-22) |
| Ripetizione parole polisillabiche | 1 (1-2) | 2 (3-4) | 3 (5) | 4 (6) |

→ **Ripetizione di liste di parole "Fonemi iniziali", "Gruppi consonantici" e "Polisillabiche"** – all n°1
Tenere conto solo del fonema/gruppo sottolineato

ALLEGATO N° 1

Ripetizione di liste di parole “Fonemi iniziali” e “Gruppi consonantici”

| | | |
|---------------|----------------|-----------------|
| <u>P</u> IPA | <u>G</u> IRO | <u>DR</u> AGO |
| <u>B</u> UCO | <u>S</u> OLE | <u>FR</u> ATE |
| <u>I</u> OPO | <u>U</u> OMO | <u>SP</u> ESA |
| <u>D</u> ADO | <u>I</u> UTA | <u>ST</u> UFA |
| <u>C</u> ASA | <u>GN</u> OMO | <u>SC</u> ALA |
| <u>G</u> ATTO | <u>GL</u> IELO | <u>SL</u> ITTA |
| <u>F</u> ILO | <u>POZZ</u> O | <u>SP</u> LENDE |
| <u>V</u> ASO | ----- | <u>STR</u> ADA |
| <u>M</u> URO | <u>BR</u> ODO | <u>SCR</u> IVE |
| <u>N</u> EVE | <u>PL</u> AGIO | <u>SPR</u> UZZO |
| <u>L</u> UNA | <u>TRE</u> NO | <u>SGR</u> IDO |
| <u>R</u> ETE | <u>CL</u> ASSE | <u>SFR</u> EGIO |
| <u>Z</u> ERO | <u>GR</u> IGIO | <u>SDR</u> AIO |
| <u>SC</u> IA | <u>FLO</u> TTA | <u>SBR</u> IGO |
| <u>C</u> IAO | <u>CR</u> ETA | <u>PR</u> OVA |

Ripetizione di parole polisillabiche

| | | |
|----------------------|-------------------|-------------------|
| <u>CALENDARIO</u> | <u>MONOTONO</u> | <u>MONTAGNOSO</u> |
| <u>AUTOBIOGRAFIA</u> | <u>PERICOLOSO</u> | <u>PRESTIGIO</u> |

“Notturmo”

Vi è un profondo silenzio, nel buio della notte.

Vicino al pozzo, nella cui acqua si specchiano la luna ed una scia di stelle, la magnolia stende i suoi rami.

Cespugli di rose olezzano nell'aria.

Il temporale è cessato, e la pioggia ormai non cade più.

Solo le rane gracidano, nei fossi, oltre quel prato.