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Research report

Progressive apraxia of speech as a window into the study of speech planning processes

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ARTICLE INFO

Article history:
Received 4 August 2010
Revised 28 September 2010
Accepted 7 March 2011
Action editor Georg Goldenberg
Published online 26 March 2011

Keywords:
Apraxia of speech
Progressive aphasia
Speech production
Syllable
Speech-motor plans

ABSTRACT

We present a 3-year follow-up study of a patient with progressive apraxia of speech (PAoS), aimed at investigating whether the theoretical organization of phonetic encoding is reflected in the progressive disruption of speech. As decreased speech rate was the most striking pattern of disruption during the first 2 years, durational analyses were carried out longitudinally on syllables excised from spontaneous, repetition and reading speech samples. The crucial result of the present study is the demonstration of an effect of syllable frequency on duration: the progressive disruption of articulation rate did not affect all syllables in the same way, but followed a gradient that was function of the frequency of use of syllable-sized motor programs. The combination of data from this case of PAoS with previous psycholinguistic and neurolinguistic data, points to a frequency organization of syllable-sized speech-motor plans. In this study we also illustrate how studying PAoS can be exploited in theoretical and clinical investigations of phonetic encoding as it represents a unique opportunity to investigate speech while it progressively disrupts.

1. Introduction

Apraxia of speech (AoS) is an impairment in speech production, which is usually ascribed to the level of planning or programming of speech gestures (corresponding to phonetic encoding in models of speech production, Levelt et al., 1999). Difficulty in accessing or generating phonetic programs results in a series of changes including phonetic and phonemic errors, groping and difficult speech initiation, changes in inter- and intra-syllabic transitions, increased syllabic duration and decreased speech rate (Code, 1998; Darley et al., 1975; McNeil et al., 2004; Varley and Whiteside, 2001). AoS has first been associated with focal brain damage (Alajouanine et al., 1939; Darley et al., 1975; Ziegler, 2005), but more recently also with neurodegenerative diseases (Duffy, 2006; Josephs et al., 2006), either in combination with progressive non-fluent aphasia (PNFA) or in isolation (Cohen et al., 1993; Didic et al., 1998; Duffy, 2006; Josephs et al., 2006; Ricci et al., 2008). In this latter case, apraxia of speech can evolve as an isolated impairment during several years without positive signs of aphasia, pointing to a pattern of pure progressive apraxia of speech (PAoS). For instance, Ricci et al. (2008) reported on a patient with atrophy in the superior frontal gyrus and isolated PAoS during 15 months. In a retrospective study on 80 patients with a clinical diagnosis of PNFA (Duffy, 2006), 11% had isolated AoS without aphasia or dysarthria, corresponding to

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http://dx.doi.org/10.1016/j.cortex.2011.03.010

a diagnosis of PAoS. This review of the literature also suggested that several previously published cases corresponded to PAoS, although different diagnostic labels were used (Duffy, 2006).

The level of impairment in AoS reflects the interface between linguistic and motor processes, i.e., the implementation of speech gestures from abstract phonological codes. One point that has received particular attention in the literature is the size and content of speech-motor programs. In one of the most influential psycholinguistic models of speech production (Levelt et al., 1999), phonetic plans are built through the activation of syllable-sized gestural scores. There is now converging evidence from the psycholinguistic and neurolinguistic literature for stored syllable-sized phonetic plans and a frequency organization of phonetic syllables. Psycholinguistic studies reported a facilitatory effect of high frequency syllables on production latencies, using different paradigms and materials (Carreiras and Perea, 2004; Cholin et al., 2006; Laganaro and Alario, 2006; Levelt and Wheeldon, 1994). Neurolinguistic studies showed that brain-damaged speakers produced more phonetic and phonemic errors on words or pseudo-words composed of low frequency syllables (Aichert and Ziegler, 2004; Laganaro, 2008; Staiger and Ziegler, 2008). Hence, stored syllable-sized motor programs are activated/accessed during phonetic encoding and those that are frequently used are better retrieved than infrequently used ones.

The patients analyzed in these neurolinguistic studies had AoS due to acquired focal brain damage. A central issue with respect to both theory and diagnostics is whether the same characteristics are observed in AoS after focal lesions and in PAoS. Actually, investigations of stroke patients are usually carried out during a stable, post-acute or chronic phase following the stroke. Consequently, at least partial recovery and some kind of reorganization may have occurred and may have influenced patients' patterns of speech production. In contrast, PAoS represents a unique opportunity to investigate phonetic encoding while it progressively disrupts.

Here we present a 3-year follow-up study of a 66-year-old man presenting with PAoS. The patient displayed progressive disruption of speech production, characterized by effortful speech, phonetic and phonemic segmental errors, inter-syllabic pauses, syllable lengthening and dysprosodia, without positive signs of aphasia. Our analyses were aimed at investigating whether the characteristics of the progressive disruption of speech in this case could shed light on the theoretical organization of phonetic encoding. As decreased speech rate was the most striking symptom during the first 2 years, acoustic analyses were carried out longitudinally on his speech samples.

Several studies have previously analyzed temporal characteristics of speech in AoS after stroke. Most of those studies carried out durational analyses on speech samples collected during word repetition and were aimed at differentiating among aphasia (especially conduction aphasia) and AoS, or to describe characteristics of AoS relative to control subjects (Collins et al., 1983; Kent and Rosenbek, 1983; Haley and Overton, 2001; Seddoh et al., 1996; Ballard et al., 2001 for a review). Unlike previous studies on durational analyses, we applied an approach usually carried out to predict error outcome. The analyses aimed at investigating which linguistic factors affect the pathological behavior. However, rather than analyzing which factors predicted error outcome, we analyzed which factors predicted the progressive disruption of articulatory rate.

A few studies on healthy control subjects have tried to track a syllable frequency effect on syllabic duration (Schweitzer and Möbius, 2004; Croot and Rastle, 2004). These investigations aimed at showing that producing speech sequences which occur less frequently is less automatic. To our knowledge, only Schweitzer and Möbius (2004) reported longer durations for very infrequent syllables than for high frequency syllables. However, these results were obtained in a post-hoc analysis on unbalanced sets of frequent and infrequent syllables and have never been replicated (Croot and Rastle, 2004). By contrast, several other linguistic variables have been reported to affect syllabic duration in studies with non-brain-damaged speakers. First, it is well established that linguistic rhythm, that is the alternation of stressed and unstressed syllables, affects syllable durations by lengthening stressed syllables compared to unstressed syllables. For instance, as the rhythmic pattern of French is iambic, words are usually stressed on their last syllables that are lengthened (Fletcher, 1991). Second, lexical frequency and phonological neighborhood density have been shown to affect articulatory properties (Pluymaekers et al., 2005; Bell et al., 2009). Bell et al. (2009) showed that word duration decreases with increased lexical frequency, especially for content words, while words with many phonological neighbors displayed hyper-articulated properties, especially if words are of low lexical frequency (Munson and Solomon, 2004; Wright, 2004).

In sum, several linguistic variables affect articulation properties and particularly syllabic duration in healthy speakers, but syllable frequency effects, which were reported on production latencies and production errors, have not been reliably reported on syllabic duration. The main goal of the present study is to test whether the theoretical organization of syllable-sized phonetic plans is reflected by progressive lengthening due to speech disruption. In other words, the question is to establish whether increasing durations are equally observed for all syllables or if disruption rather follows the ease to retrieve and execute stored motor plans. The first hypothesis predicts that all syllables should undergo a constant increase in duration. The second hypothesis predicts variable lengthening across syllables. In that case, lengthening should be smaller for high frequency syllable-sized motor plans.

2. Method

2.1. Case description

The case presented in this study is that of a 66-year-old man with 12 years of education, retired precision mechanic and an amateur ventriloquist. More than 1 year before the first assessment he realized that he was having difficulties in ventriloquism and in playing the accordion. In a consult, at the age of 66, he complained about difficulties in speech production. A detailed neuropsychological test battery was carried out at first examination (January 2007) and repeated 20 months (November 2008) and 34 months later (December 2009). Speech samples were collected at the same dates.

2.2. First examination

The first neuropsychological examination (January 2007) revealed no signs of aphasia, with performance within the normal range in other cognitive domains, except for mild executive dysfunction (see Table 1). Speech rate was mildly slow with occasional phonetic errors and reduced diadochokinetic rate. Singing was possible but the patient could no longer play ventriloquism (normally visible – not posteriorized – speech was produced when trying ventriloquism).

The first neurological examinations (January 2007, March and May 2007) were within normal range except for a very slight loss of hand dexterity and minimal finger tapping hypokinesia on the right.

In 2007, at the time of the first assessment, brain magnetic resonance imaging (MRI) and FDG–PET scans revealed atrophy and hypometabolism respectively of the left insula, the left inferior, medial and superior frontal gyrus and the left precentral gyrus (see Fig. 1).

2.3. Second examination (20 months later)

At second assessment 20 months later, speech rate had drastically decreased, with an increase in phonetic and phonemic errors (see analyses below). His neuropsychological profile was virtually unchanged (see Table 1), except for mild behavioral changes (reduced social interactions and irritability). He also displayed increased articulation difficulties with longer response latencies.

The neurological examination at that time revealed dystonic posture, extrapyramidal rigidity and hypokinesia of the right hand, as well as decreased right arm swing when walking.

2.4. Third examination (34 months later)

Three years later, at third assessment, speech rate was severely reduced with many latencies due to both inter- and intrasyllabic pauses and word finding difficulties. Only at this assessment point anomia was observed and verbal fluency was severely impaired (see Table 1). During the last 12 months motor difficulties and cognitive impairment have also aggravated.

At that time, a neurological exam disclosed a full corticobasal syndrome characterized by dystonic posture of the right hand with extrapyramidal rigidity, marked bradykinesia, hand levitation, synkinesia, grasping behavior, hypesthesia and alien hand syndrome, as well as a very disturbed gait due to the extrapyramidal syndrome. Limb apraxia was bilateral but predominant on the right and particularly on imitation of intransitive non-representational gestures.

In sum, the patient presented with isolated/predominant PAoS during at least 20 months. He displayed progressive disruption of speech production, characterized by phonetic

Table 1 – Results of neuropsychological assessment at each assessment period.

	January 2007	November 2008	December 2009
Speech	Mildly slowed, reduced diadochokinetic rate	Severely reduced speech rate, phonetic/phonemic errors	Severely reduced speech rate, phonetic/phonemic errors, echolalia
Language			
Boston naming, French version (Colombo and Assal, 1992)	NR	NR	Mild impairment
Action naming, DVL38 (Hammelrath, 2001)	NR	NR	NR
Compréhension (Nespoulous et al., 1992)	NR	NR	NR
Writing	NR	NR	Motor difficulties and some letter inversions
Calculation	NR	NR	NR
Semantics			
PPT (Howard and Patterson, 1992), Lexis (De Partz et al., 2001)	NR	NR	NR
Memory			
Verbal Span	Limit	Limit	Mild impairment
Grober and Buschke, 1987 (RL-RI 16)	NR	NR	NR
Complex figure (Osterrieth, 1944)	NR	NR	n.a.
Executive functions			
Verbal fluency	Mild impairment	Mild impairment	Severe impairment
Stroop, Kramer	Mild impairment	Mild impairment	Severe impairment
Attention	NR	NR	NR
Visual perception	NR	NR	NR
Gesture			
Limb	NR	Right hand slow, no apraxic errors	Apraxic
Oral	Slow, no apraxic errors	Slow, no apraxic errors	Apraxic

NR: normal range. n.a.: not assessed.

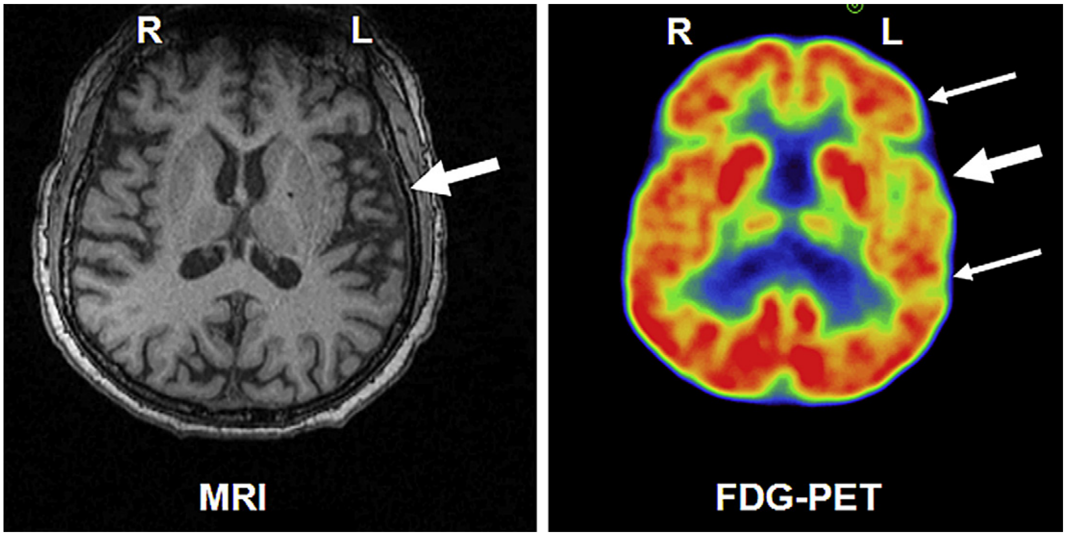

Fig. 1 – Axial MRI scan and corresponding FDG–PET scan respectively showing atrophy and hypometabolism of the insula (thick arrows) with hypometabolism extending both anteriorly and posteriorly to the atrophy (thin arrows).

and phonemic errors, inter-syllabic pauses, syllable lengthening and dysprosodia, without positive signs of aphasia during the first two assessments. Only at third assessment (34 months later) did positive signs of aphasia appear, together with severely reduced speech rate, echolalia and limb apraxia.

2.5. *Speech sample collection and pre-analyses*

Speech samples were collected and recorded during natural conversation, word/sentence repetition and sentence reading. All speech samples were digitized, transcribed and analyzed using the speech analysis software Praat (Boersma and Weenik, 2007) by two independent trained judges. Error rate and syllable duration were analyzed at each of the examination phases, i.e., at first assessment (January 2007), 20 months later (November 2008) and 34 months later (December 2009).

Errors were auditorily perceptible phonetic and/or phonemic segmental transformations. Acoustic analyses were conducted by combining auditory judgment and visual inspection of speech waveforms and spectrograms. Syllable boundaries were identified and labeled using standard segmentation criteria, with labels placed at the point of zero crossing on the waveform. Initial and final vowel boundaries were respectively determined by identifying the onset/the end of a visible pitch period that corresponds to a regular formant structure. Initial fricative boundaries, were placed at the onset of visible frication noises. For nasals and liquids in syllable onsets, a visible change in the distribution of energy (e.g., nasal formants) was taken as the initial segment boundary. For voiced stops, initial boundaries were placed at the beginning of the voice bar produced during the closure. For unvoiced stops, the beginning of a silent period was used as an indicator of the onset of the consonant. Final boundaries were placed after the end of frication noises, nasal formants or burst created by the release of the closure, respectively for fricative, nasal and stop consonants.

Inter-judge agreement on syllabic duration was assessed on a sample of 120 commonly analyzed syllables from spontaneous speech at first and second assessment. Pearson correlation on durations across the two judges was .917; 83% of the absolute difference in syllabic duration across judges was below 50 msec (below 10% of mean syllabic duration).

2.6. *Analyses*

To test the hypotheses on factors affecting syllable lengthening, we carried out a set of analyses on duration of CV syllables only. These analyses were limited to CV syllables for the following reasons. First, syllabic duration could not be compared across different syllabic structures as they varied in the number of phonemes. Second, CV syllables are the most frequent syllables in French and in the collected speech sample (58.4% in the entire corpus), ensuring enough measures when same syllables in identical word position were analyzed across recording sessions.

Besides syllable frequency, the following factors were considered, as they are known to modulate syllable duration (see the Introduction): *position* of syllable in word, *word length*, *lexical frequency* and *phonological neighborhood density*. *Bi-phone frequency*, *phoneme frequency* and the *frequency of the following syllable* were also considered in the analyses, as they represent possible confound factors of a syllable frequency effect. All lexical and sub-lexical properties were taken from the French database LEXIQUE (New et al., 2004). All frequency measures were log transformed for the following analyses.

As a set of analyses was carried out on the entire data with different statistical approaches, each analysis will be detailed in the corresponding result section.

3. Results

3.1. *Errors*

Table 2 summarizes the sample size and the number of errors.

All errors were phonetic or phonemic errors including distortions and phoneme substitutions, omissions and *schwa*

Table 2 – Analyzed speech sample (total N. of syllables) and percent errors.

	First assessment (February 2007)	20 months later (November 2008)	34 months later (November 2009)
Total number of syllables	430	278	536
Spontaneous	203	86	227
Repetition (sentences and words)	75	111	209
Reading (sentences)	152	81	100
Errors (per syllable)	1.4%	4.7%	6.9%

or vowel insertion (ex. *Silvie* (/silvi/) produced [silivi], *privilège* (/pRivilƐƷ/) produced [fRivilƐƷ], *oreille* (/oRƐj/, ear) produced [aRƐj]). Error rate increased slightly but significantly from first to second assessment (Pearson chi-square, $\chi^2(1) = 6.96, p < .01$) while the difference did not reach significance between the second and third assessments ($\chi^2(1) = 2.16, p < .13$). Despite an increase in phonetic and phonemic segmental transformation, the total number of errors did not allow further analysis relative to syllable frequency.

3.2. *Speech rate and articulation rate (syllable duration)*

Speech rate (number of syllables/sec, including filled and empty pauses) was already slow in 2007 and strikingly decreased at the following assessment periods (see Table 3), with many intra-clause and intra-syllabic pauses.

Articulation rate (number of syllables/sec, excluding pauses and inter-syllabic transitional segments) substantially decreased from first to second assessment and only slightly at third assessment. Decrease of articulation rate was noted for all production modalities to the same degree. Comparison on articulation rate between the three assessment periods was carried out on duration of identical V, CV and CVC syllables. On these identical syllables ($N = 65$) duration was almost doubled at second examination, across all syllabic structures (Table 3) and slightly decreased at third assessment. A repeated measures analysis of variance (ANOVA) across the three assessment sessions indicated a significant effect of session on syllabic duration [$F(2, 64) = 186{,}05$, $p < .0001$]. At planned comparisons, all between-session differences were significant (Fisher test, all $p < .001$).

3.3. *Factors affecting syllable duration*

The first analysis was carried out on all CV syllables. A multiple stepwise linear regression was run separately for each assessment session with syllabic duration as dependent variable and lexical and sub-lexical predictors.

In order to accurately select the factors to be entered in the multiple regression analysis, we visualized the collinearity structure of our predictors with hierarchical clustering. To address the collinearity structure of possible predictors, we followed the procedure suggested by Baayen (2008, p. 198–201). Then, given the collinearity between several variables we applied the simplest strategy of first entering only one factor from each cluster in the model, but we systematically also tested the other variables from each cluster. This means that the initial model had the three following factors: *syllable frequency*, *position in word* and *lexical frequency*. We then systematically replaced lexical frequency with the other factors in the cluster (with *phonological neighborhood* first, then with *word length*). The best model for each session was selected based on likelihood ratio statistics and on the residual standard errors following the procedure suggested by Baayen (2008).

The overall regression models were significant [2007: $F(3, 244) = 3.24$, $p < .05$, $R^2 = .040$; 2008: $F(3, 151) = 13.51$, $p < .0001$, $R^2 = .212$ and 2009: $F(3, 314) = 9.17$, $p < .0001$, $R^2 = .081$]. A summary of the models is presented in Table 4.

Position was the common predictor of CV syllable duration in all assessment periods: syllabic duration increased when the syllable was at the end of the word in the 2007 and 2008 data, but this effect was in the opposite direction (decreased with position in word) at last assessment.

For the other factors, the models differed between 2007 and the two following assessments. In 2007, phonological neighborhood density was the main predictor of duration (notice however the small R^2 for the 2007 model). In 2008 and 2009, lexical frequency and syllable frequency predicted CV syllable duration. These two effects had opposite directions: syllables had longer duration when contained in words with high lexical frequency, but had a shorter duration for high frequency syllables. Thus, an opposite effect of lexical frequency and of

Table 3 – Speech rate, articulation rate (syllables/sec) and mean syllable duration (in msec) for common syllables at each assessment period.

	2007	2008	2009
Speech rate (syll/sec)			
Spontaneous	1.78	1.28	.74
Articulation rate (syll/sec)			
Spontaneous	2.70	1.68	1.56
Repetition	3.03	1.67	1.51
Reading	2.92	1.79	1.49
Syllable duration (msec)			
V	214	438	486
CV	329	518	605
CVC	397	731	833

Table 4 – Multiple regression analysis on the duration of CV syllables.

	Predictor	β	t	p
2007	Position	.135	1.82	.07
	Neighborhood	.231	3.07	.002
	Syllable frequency	−.070	−1.10	>.1
2008	Position	.247	2.989	.003
	Lexical frequency	.364	4.453	.000
	Syllable frequency	−.373	−4.854	.000
2009	Position	−.118	−2.081	.038
	Lexical frequency	.187	3.304	.001
	Syllable frequency	−.144	−2.652	.008

syllable frequency on duration was observed when articulation rate decreased. Crucially, syllable frequency correlated with duration in each production modality (spontaneous, reading and repetition in 2008, see Appendix).

Before any further analysis, we first had to exclude any possible confound of the observed effects. *Lexical frequency* differs across content words and function words (the latter are usually mono- or di-syllabic and are high frequency words). Therefore, longer syllabic duration for high frequency words might be linked to pathological lengthening limited to function words. When only content words were analyzed, results replicated those reported above, whereas on function words only syllable frequency was a significant predictor of syllabic duration in 2008 (Beta $= -.337$, $t = -2.34$, $p < .05$) with no further significant factors in 2008 and no significant predictors for closed class words in the other assessment sessions.

The correlation between lexical frequency and neighborhood density might reflect a possible confound between these two factors, which should affect duration in opposite directions (see Introduction). Further exploration of the data split between words with many phonological neighbors and words with few neighbors indicated positive correlation between lexical frequency and duration only for words with many phonological neighbors (respectively: $r = .362$ and $r = .233$ in the 2008 and 2009 data); by contrast the correlation was negative in 2007 ($r = -.299$). When the opposite splitting (low and high frequency words) was applied to the data, a positive correlation was observed between neighborhood density and duration in the 2007 data ($r = .222$).

Concerning syllable frequency, other factors like phoneme frequency or bi-phone frequency might carry the observed effect. Alternatively, CV syllables might be lengthened in order to plan a following complex syllable; in this case the frequency of the following syllable might predict duration.

We entered each of these factors (phoneme frequency, bi-phone frequency of the analyzed syllable and the frequency of the following syllable) one by one in the previous models by replacing syllable frequency and compared those models to the original models on likelihood ratio statistics and the variability in the residuals. For the 2007 data the model fitted better with phoneme frequency, approaching significance (Beta coefficient: $-.135$, $t = -1.88$, $p = .06$). By contrast, the model with syllable frequency had a better fit over all other models for the 2008 and 2009 data. We can therefore be confident that syllable frequency predicted duration in the two last assessments.

3.4. *Predictors of difference in duration across assessment periods*

In order to further test whether syllable frequency affected the progressive lengthening of syllabic duration, we analyzed which factors predict the difference in duration between consecutive assessment sessions. We therefore considered only CV syllables which appeared in all assessment sessions. As word position effects were observed across assessment periods, only common syllables in same position in words of same length were considered ($N = 37$). Difference in duration was computed by subtracting the mean duration of identical syllables in identical word position from the previous assessment from the duration of the following assessment.

Mean durational differences for these common syllables between 2007 and 2008 were 199 msec (SD $= 134$ msec). There was a significant correlation between difference in duration and syllable frequency ($r = -.411$, $N = 37$, $z = -2.55$, $p = .01$), indicating smaller lengthening for high frequency syllables. Mean difference on common syllables in same position between 2009 and 2007 was 253 msec (SD $= 127$ msec), with no significant correlation with syllable frequency ($r = -.229$, $z = -1.36$, $p > .1$). Importantly, the syllable frequency effect on lengthening between 2007 and 2008 was not accounted for by differences in lexical frequency of the carrying words, as there was no correlation between difference in duration and difference in lexical frequency ($r = .112$, $N = 37$, $z < 1$).

4. Discussion

The 3-year follow-up study of this patient revealed a progressive decrease of speech rate characterized by syllabic and inter-syllabic pauses lengthening, accompanied by an increase of phonetic and phonemic segmental errors. Together with the perceptible impression of effortful speech, dysprosody and initiation latencies in the absence of positive signs of aphasia, this pattern corresponds to PAoS at least during the first 20 months. Only at third assessment (34 months after first examination), did neuropsychological assessment also disclose positive signs of aphasia and decline in other cognitive domains as well as motor disease. The pattern of isolated PAoS during the first 2 years, followed by motor and cognitive decline, matches similar case descriptions in the literature (Duffy, 2006; Ricci et al., 2008); in most cases AoS was associated with corticobasal degeneration or supranuclear palsy (Josephs et al., 2006), the former being the probable underlying neuropathology in the present case.

Our main aim here was to investigate the organization of speech-motor plans through the analyses of disruption of articulatory rate (of syllabic duration). We analyzed longitudinally CV syllable duration and assessed whether syllable duration increased according to a fixed factor across all syllables or if it reflected the frequency of use of syllable-sized motor programs.

The main result thus concerns the factors affecting the striking increase in syllabic duration during speech disruption. While word position and lexical effects were observed at first examination and were still present in 2008 when speech rate drastically decreased, syllable frequency predicted syllable lengthening at second and third assessment. This effect was present independently of the eliciting task and was corroborated by an effect of syllable frequency on difference in syllabic duration for same syllables across the first and the second assessment periods.

We will shortly discuss the effect of lexical variables on syllable duration first, and then turn to our main result (syllable frequency).

4.1. *Lexical effects on duration*

The positive correlation between syllable duration and position within the word indicates that normal lengthening for final (stressed) syllables in French (Fletcher, 1991) is preserved despite severe reduced articulatory rate, at least in 2008. This was no longer the case at last assessment, when speech rate further decreased and more general decline was noticeable.

The effect of lexical factors (phonological neighborhood and lexical frequency) at all assessment sessions also seems to indicate preservation of normally observed patterns of syllable duration. At first glance, the relationship between lexical frequency and syllable duration was in the opposite direction to those reported in studies with healthy speakers, in which frequent words have rather been reported to have reduced phonetic properties (Pluymaekers et al., 2005; Bell et al., 2009). However, split between words with many phonological neighbors and words with few neighbors indicated an interaction between those two factors. These two factors were not considered jointly in previous studies (nor was syllable frequency), we can therefore not conclude about the direction of those factors relative to previous reports.

4.2. *Syllable frequency effects on duration*

The central result of the present study is the demonstration of an effect of syllable frequency on syllabic duration. Syllable frequency affected syllabic duration independently of other effects and after many possible lexical and sub-lexical confounds (lexical frequency, phonological neighborhood, position in word, phoneme and bi-phone frequency and the frequency of the following syllable) were eliminated. Although other possible confounds at lexical and syntactic level might affect syllable duration, the consistency of results across production tasks (reading, repetition and spontaneous production) and across content and function words make us very confident that syllable frequency played a role in the disruption of speech in this case of PAoS. Syllable frequency predicted syllabic duration at second and third assessment; moreover, syllable frequency correlated with the difference in duration of identical syllables between the first and the second recording sessions.

These results seem to indicate that impaired phonetic encoding does not affect all phonetic plans in the same way, but respects a gradient which is a function of the frequency of use of the motor programs. The present results also suggest that lengthening is not a mere compensation mechanism. If this was the case all syllables should have been affected to the same extent as a function of the upcoming articulatory difficulty (the difficulty of the following syllable did not affect syllabic duration).

Previous studies investigating AoS after stroke have already reported an effect of syllable frequency on accuracy, with more phonetic and phonemic errors on words composed of low frequency syllables (as discussed in the Introduction). The present data point toward an additional effect of syllable frequency on pathological lengthening of syllabic duration. Importantly, in studies reporting error analyses, syllabic structure (syllabic complexity or syllabic constituents) also affected error rate in patients with AoS (Romani and Galluzzi, 2005; Ziegler, 2005, 2009), creating a possible confound of syllable frequency effects, as complex syllables correspond to less frequent syllables. This effect was controlled for in the present study as only CV syllables were analyzed.

We are not aware of any previous investigation analyzing syllable frequency effects on syllabic duration in AoS, and the studies on healthy control subjects reviewed in the Introduction have led to inconsistent results. It appears that in normal production, frequently used motor programs can be accessed faster than infrequently used ones (as attested by reaction time studies, discussed in the Introduction), but there is no reliable evidence that they are articulated faster. In speech pathology, frequent syllables are more resistant to errors (Aichert and Ziegler, 2004; Laganaro, 2005, 2008; Staiger and Ziegler, 2008) and to pathological lengthening. Two possible reasons can account for this effect on pathological increased syllabic duration.

First, stored syllabic motor programs may become inaccessible in case of impaired phonetic encoding, requiring the assembling of phonemes or other sub-syllabic units (Varley and Whiteside, 2001). The production of inaccessible/un-stored motor plans would therefore involve longer duration due to reduced intra-syllabic co-articulation. However, this hypothesis predicts a categorical difference between stored and inaccessible/un-stored motor-speech programs rather than a gradient in function of syllable frequency.

Second, as hypothesized in the studies investigating durational effects in healthy participants, co-articulation is harder for less frequent speech-motor plans, leading to lengthening of infrequent syllables. A possible reason why this hypothesis was not confirmed on normal speech production is that differences across syllables of high and low frequency are not great enough to be captured in normal conditions independently of other lexical and sub-lexical factors affecting syllabic duration. By contrast, when phonetic encoding gets impaired as in PAoS, the increased difficulty in preparing motor plans lengthens durations and therefore differences could be captured in the present study. This hypothesis predicts that all stored phonetic representations are affected and the resistance of phonetic plans to disruption depends on their frequency of use. Therefore, the increased cost in accessing or implementing syllabic plans reflects the organization of syllable-sized motor programs.

In conclusion, the progressive disruption of syllabic duration in PAoS follows a gradient that is a function of the frequency of use of syllable-sized motor programs. This finding adds additional evidence to a frequency organization of syllable-sized phonetic plans. It also illustrates how the analysis of pathological speech in PAoS can be exploited for theoretical and clinical investigations of phonetic encoding.

Acknowledgments

This research was conducted with the support of Swiss National Science Foundation (grants #PP01-118969 and #CM30-124115) and of Private Funding from the Société Académique de Genève (Fonds Cognitive Memory 2008/70). We are also very grateful to Marian van der Meulen for her careful proofreading of the manuscript.

Appendix

Correlations between syllabic duration and lexical and sub-lexical factors for each assessment period and task.

	Position in word	Word length	Lexical Frequency	Neighbor density	Syllable frequency
2007					
All tasks: $N = 249$	−.004	**−.155***	.094	**.156***	−.032
Spontaneous (137)	−.048	**−.168***	.120	**.184***	−.109
Repetition (29)	−.103	.219	.235	.325	.405
Reading (84)	**.218***	.039	−.086	.013	−.055
2008					
All tasks: $N = 155$	.023	−.131	**.270****	.121	**−.320***
Spontaneous (44)	−.224	**−.408****	.292*	.169	**−.447****
Repetition (59)	.149	.058	**.351****	−.079	**−.330****
Reading (52)	.027	−.223	.200	.099	**−.289***
2009					
All tasks: $N = 319$	**−.178****	−.097	**.177****	.124	**−.142****
Spontaneous (143)	**−.250****	**−.191***	**.233****	**.231***	−.061
Repetition (115)	−.123	.009	.140	.081	−**.174***
Reading (60)	−.024	.068	.075	−.086	**−.239(*)**

$^* = p < .05$, $^{**} = p < .01$, (*) = $p = .06$. Significant correlations are in bold.

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