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Changes in lingual-alveolar contact pressure during speech over six months in amyotrophic lateral sclerosis

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ABSTRACT

Purpose: This study compared lingual-alveolar contact pressures during consonant (LACP-Sp) production between adults with and without amyotrophic lateral sclerosis (ALS) over three visits within six months and assessed the relationship between LACP-Sp and speech intelligibility.
Methods: Ten people with ALS and nine without read sentences with lingual-alveolar consonants embedded as LACP-Sp was recorded. Recordings were done at study entry, then three and six months after entry. Six of ten with ALS had bulbar symptoms detected in clinical encounter at the time of study enrollment (ALS-B); the remaining four had only spinal symptoms (ALS-S).
Results: Main and interaction effects varied across experimental stimuli. However, the pattern of results from the ANOVAs indicated that the ALS-B had lower LACP-Sp values than the other groups. Time main effects were present indicating a reduction in LACP-Sp. However, there were interaction effects for fricatives and/l/, with the ALS-S group demonstrating greater reduction over time than the other groups. Word intelligibility had moderate to large correlations with LACP-Sp but confidence intervals were broad and only a few were statistically significant.
Conclusion: LACP-Sp does decrease over time in people with ALS and may reflect a general weakening of the tongue. Confirmation of these group changes will require assessment with a larger sample size.

1. Introduction

A primary feature of amyotrophic lateral sclerosis (ALS) is the progressive nature of the disease. The progression is often rapid with the rate of decline associated with a variety of factors such as site of symptom onset in the body, gender, and age, among other variables (Czaplinski, Yen, Simpson, & Appel, 2006; Wolf et al., 2014, 2015). Those with symptom onset in the head and neck (bulbar) comprise about 30% of the individuals diagnosed with ALS and they have consistently been found to have shorter survival times than those with symptom onset in the limbs (Moura et al., 2015; Pupillo, Messina, Logroscino, & Beghi, 2014). Regardless of the site of onset, both upper and lower motor neuron symptoms can occur including muscle weakness, spasticity, fasciculation, hyperreflexia, and atrophy, among other symptoms (Mancuso & Navarro, 2015). When ALS impacts bulbar musculature, dysarthria and dysphagia will eventually become apparent for the majority of patients (Beukelman, Fager, & Nordness, 2011; Strand, Miller, Yorkston, & Hillel, 1996).

The progressive nature of ALS and short survival times, particularly for those with bulbar symptoms, has prompted researchers and clinicians to identify measures that are sensitive to symptom onset and change over time. The search for valid, reliable, and

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clinically feasible means of detecting the onset of bulbar symptoms is important for various reasons including the ability to select and stratify patients for clinical trials of pharmaceuticals or other interventions, to serve as an outcome measure in such trials, and to guide clinical decision making if a measure is a strong predictor of impending change in disease state or function. Relative to detection and tracking of bulbar symptoms, a variety of approaches have been investigated. For example, a reduction in speaking rate below about 120 words per minute has been identified as an antecedent to a precipitous drop in speech intelligibility in adults with ALS (Yorkston, Strand, Miller, Hillel, & Smith, 1993). However, Mefferd, Green, and Pattee (2012) have reported articulatory kinematic data indicating that individuals with very mild articulatory weakness from ALS can adjust speech motor movements to maintain speaking rates that are considered to be within a normal range. As such, speaking rate may not be sensitive enough to the earliest onset of bulbar weakness in ALS. Rong and colleagues (Rong, Yunusova, Wang, & Green, 2015) have provided a brief summary of several other potential quantitative measures across speech subsystems (respiratory, laryngeal, resonatory, oral articulatory) that have been investigated over the years. These include a range of measures such as vital capacity (VC), maximum ventilatory volume, fundamental frequency (F0) variability, restricted F0 range and a host of other acoustic measures, nasal air escape, and speed and distance measures of articulatory movements. Given that the clinical presentation of bulbar symptoms can vary markedly across patients with ALS it is not likely that a measure reflective of a single speech subsystem will be adequate for devising robust diagnostic procedures to detect early disease onset and sensitively track changes in symptoms over time. Therefore, the call for an approach that considers function across all speech subsystems has high merit (Green et al., 2013; Rong et al., 2015; Yunusova, Green, Wang, Pattee, & Zinman, 2011).

A multi-system approach, however, requires identification of specific measures within each of the speech subsystems. The work by Green and colleagues has identified several candidate measures with strong predictive value including VC, subglottal air pressure, F0, maximum phonation duration, nasalance, and kinematic measures of the lips, tongue, and jaw, among several other measures (Rong et al., 2016; Yunusova et al., 2010, 2012; Yunusova, Green, Lindstrom, Pattee, & Zinman, 2013). Recently, preliminary work on a measure of the contact pressures generated by the anterior tongue on the palate during production of lingual-alveolar consonants in adults with ALS was reported (Searl, Knollhoff, & Barohn, 2017) (lingual-alveolar contact pressure during speech [LACP-Sp]). It is not yet known if this could potentially be of use in a multi-system measurement approach. The instrumentation and procedures for obtaining such contact pressure measurements during speech production have been reported for adults without speech disturbance (Searl & Evitts, 2013; Searl, 2003; Searl, Evitts, & Davis, 2007) and for describing articulatory changes after total laryngectomy (Searl, 2007, 2008). The recent application to individuals with ALS was intended to investigate the feasibility of the obtaining LACP-Sp in this clinical population and to gather initial data about differences in speech contact pressures between adults with and without ALS. Those preliminary results indicated that adults with ALS presenting with bulbar symptoms had significantly lower contact pressures during lingual-alveolar stops and fricatives compared to individuals with ALS who had spinal symptoms and compared to adults without ALS. One tentative interpretation of those data was that LACP-Sp may be reflective of bulbar weakness in the tongue and ability to document such weakness during speech production (rather than non-speech pressing) could help elucidate the role of declining strength on speech production in ALS. Several questions about LACP-Sp remain, however, before serious consideration can be given to pursuing the measure more aggressively.

One basic question regarding the measure of LACP-Sp is how stable the values are over time in a person who is considered to have normal speech. Although LACP-Sp has been measured in non-disordered adults previously, there has not been demonstration of the longer-term stability of the measure. If LACP-Sp varies markedly within a person across recording days, weeks, or months then its utility for any purpose is restricted or perhaps nonexistent. The first aim of this study was to evaluate LACP-Sp for a group of adults without ALS at three recording times within a six-month interval. A second aim was to compare LACP-Sp over a six-month interval for individuals with ALS to those without ALS. Stability in the non-ALS group and reduction over time in the ALS participants would highlight the possibility that LACP-Sp was reflective of a change in disease state in the ALS group. Although the number of participants in the current study is small, roughly half had ALS symptoms only in the limbs (spinal symptoms [ALS-S]) and the other half had already detectable bulbar symptoms (ALS-B) allowing some initial consideration of disease state relative to LACP-Sp. Finally, the strength of the relationship between the physiological measurement, LACP-Sp, and word intelligibility was assessed. Absence of a strong relationship would dampen interest in further pursuing the physiological measure if it did not appear to be relevant to a more global measure such as intelligibility.

2. Materials and methods

The participants, instrumentation, and procedures are taken from Searl et al. (2017) and extended here to include measurements at three and six months after the initial recording. Additional description of the participants is offered below because the group became smaller over the course of this longitudinal study.

2.1. Participants

All participants provided written consent and the study was approved by the IRB committee at the University of Kansas Medical Center. Six participants in the original report are not included here: three with ALS passed away during the study and three without ALS withdrew for unspecified reasons after the baseline visit. This left 10 individuals with ALS and 9 without who participated in the current study. Table 1 includes demographic and medical history information for the participants. The individuals with ALS were subdivided into those with clinically documented bulbar symptoms at time of consent (ALS-B) and those who had only spinal bulbar symptoms at time of consent (ALS-S) based on the clinical notes from the treating neurologist and SLP. The ALS-B group consisted of

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