

Speech-associated Labiomandibular Movement in Mandarin-Speaking Children with Quadriplegic Cerebral Palsy: a kinematic study

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Abstract (200 words)

The purpose of this study was to investigate the speech-associated labiomandibular movement during articulation production in Mandarin-speaking children with spastic quadriplegic (SQ) cerebral palsy (CP). Twelve children with SQ CP (aged 7 to 11 years) and 12 age-matched healthy children as controls were enrolled for the study. All children underwent analysis of percentage of consonants correct (PCC) and kinematic analysis of speech tasks using the Vicon Motion 370 system. Kinematic parameters included utterance duration, displacement and velocity of the lip and jaw, coefficient of variation (CV) of lip utterance duration, and spatial and temporal coupling of labiomandibular movement of speech produced in mono-syllable (MS) and poly-syllable (PS) tasks. Children with CP showed lower temporal coupling (MS, $p=0.015$; PS, $p=0.007$), but not spatial coupling, of labiomandibular movement than healthy children. Children with CP had greater CVs (MS, $p=0.003$; PS, $p=0.010$) and the peak opening displacement and velocity of lower lip and jaw ($p<0.05$), and lower PCC ($p<0.001$) than healthy children. Children with SQ CP displayed labiomandibular coupling movement impairment, especially in the aspect of temporal coupling. These children also had high temporal motor variability and needed to make more effort to coordinate the labiomandibular movement for speech production.

Keywords: Speech; Cerebral palsy; Kinematics; Labiomandibular Movement; Speech intelligibility

1. Introduction

Cerebral palsy (CP) describes a group of disorders in development of movement and posture, which often causes activity limitation and may be attributed to non-progressive disturbances occurring in developing fetal or infant brains (Rosenbaum et al., 2007). CP occurs approximately 0.1-0.2 % of births each year, and serves as one of the most common motor disorders among children (Paneth & Hong, 2006). Spastic quadriplegic (SQ) CP accounts for 10-15% of spastic CP (Singhi, Ray, & Suri, 2002) and is the most severe form of spastic CP. Children with SQ CP often suffer swallowing, speaking, and intellectual problems (Straub & Obrzut, 2009). They possess lower speech functions than those with spastic diplegia (Chen et al., 2010a; Lee et al., 2010).

Children with CP usually show speech impairments (Chen et al., 2010b). Poor speech production may result directly from disturbed neuromuscular control of speech mechanisms (Pirila et al., 2007). The motor impairment severity, cognitive functions (Chen et al., 2010b; Parkes, Hill, Platt, & Donnelly, 2010), and language communications are associated with the motor speech control among children with CP (Chen et al., 2010a; Chen et al., 2010b). For example, children with spastic CP commonly exhibit dysarthria of varying severities (Marchant, McAuliffe, & Huckabee, 2008). Reduced intelligibility can adversely impact communication abilities and limit their vocational, educational, and social participation (Hustad, 2008). Speech impairments also are at risk for limited literacy development in children with CP (Peeters, Verhoeven, de Moor, & Balkom, 2009).

Speech movement coordination requires the interaction of speech articulators into larger functional aggregates (Gracco & Löfqvist, 1994). These articulatory aggregates are the framework for speech motor control and their activation is associated with sound production (Gracco & Löfqvist, 1994). Normal speech development involves the integration of lip and tongue activities into a more well-established dominant jaw operating sensorimotor system (Dworkin, Meleca, & Stachler, 2003). In contrast, mandibular dyscontrol may account for articulation deficits in children and adults with developmental or neurogenic disorders (Dworkin et al., 2003; Green, Moore, & Reilly, 2002). Previous studies have employed peak coefficients and lag-to-peak coefficients derived from the cross-correlation functions to measure the spatial aspects of articulatory movement (spatial coupling) and the degree of movement synchrony (temporal coupling) between articulatory pairs (e.g. lower lip to jaw) (Green, Moore, Higashikawa, & Steeve, 2000). Analysis of the labiomandibular coupling movement should also provide insight into the speech movement coordination process. However, no research up to date has investigated the labiomandibular coupling movements in children with spastic CP using a kinematic analysis.

Treating motor speech dysfunction in children with SQ CP requires an understanding of the

mechanism underlying speech motor control (Chen et al., 2010c). A previous study that applied kinematic analysis to investigating speech motor control in children with mild CP has shown that high spatiotemporal index values reflect deficits in relative spatial and temporal control for speech in the CP children (Chen et al., 2010c). However, it remained unclear how the labiomandibular movements are controlled and coordinated for speech production in children with SQ CP. We hypothesize that the labiomandibular coupling movement and oromotor variability during speech was impaired in children with SQ CP due to deficits in oromotor control. This study aims to investigate the control and coordination of speech-associated labiomandibular movement in Mandarin-speaking children with SQ CP using a kinematic analysis. The kinematic parameters used to detect speech motor control problems in the present study may potentially have practical clinical applications.

2.Methods

2.1. Participants

Twelve children with SQ CP (7 males, 5 females), aged 7 to 11 years old (8.9 ± 1.6 years) from the rehabilitation department of Chang-Gung Memorial Hospital, a tertiary hospital, were enrolled in this study. The inclusion criteria were as follows: (1) CP with spastic quadriplegia with articulation disorders; (2) good cooperation during examination; and (3) ability to understand verbal commands and perform the tasks required for this study. Exclusion criteria included any history of the following within the previous three months: (1) significant medical problems such as active pneumonia; (2) significant hearing impairment; (3) major surgical treatment, such as orthopedic surgery; (4) treatment involving nerve block, such as a botulinum toxin injection; and (5) history of facial palsy. The control group consisted of 12 age-matched healthy children (7 males and 5 females) aged 7 to 11 years old (8.6 ± 1.6 years) with no history of learning disabilities, speech or language impairments, neurological lesions, or visual or hearing impairments. Their speech functions were screened by a speech pathologist. This study was approved by the local medical ethics and the human clinical trial committee at Chang Gung Memorial hospital, and all participants gave informed consent signed by both participants and their guardians.

2.2. Instrumentation

A six-camera motion-analysis system (VICON 370, Oxford Metrics Inc) incorporating a digital camcorder was used to capture the movement of reflective markers attached to a child's mask and face and to video images during speech tasks. For each speech task, a hand-controlled digital signal synchronized with an external LED light was used to synchronize the kinematic data with the video and determine onset and offset of marker movement. Kinematic data of the reflective markers were recorded at a sampling rate of 60Hz and digitally low-pass filtered using a bi-directional zero phase-lag Butterworth filter with 5Hz cut-off frequency. The 5-Hz cut-off frequency was determined as optimal by residual analysis

suggested by Winter (1990) and was used to reduce markers' velocity error, which might be introduced by noise signal using numerical differentiation method, without significantly altering the results of marker displacements.

2.3. Assessment Procedures

We analyzed speech intelligibility and performed kinematic analysis of speech tasks on all children.

2.3.1. Speech intelligibility assessment

The percentage of consonants correct (PCC) task, modified from procedures outlined by Shriberg & Kwiatkowski (1982), was used to determine the severity of speech intelligibility. The procedures for speech intelligibility measurement was described in our previous study (Chen et al., 2010c). The task utilized 140 word cards for school children. Each subject was seated in a quiet room where an examiner showed him or her word cards in a predetermined order. The subject was then asked to read each card aloud in a normal voice. For each unfamiliar word, the examiner either explained the word or asked the subject to read it with the assistance of a phonetic transcription. The subject was told that the words he or she read were being recorded. The examiner then recorded a speech sample of the subject.

To measure PCC, a rater must make correct-incorrect judgments of individual sounds produced in the speech sample of each subject. The rater, who is a native Mandarin speaker with normal hearing, transcribed recorded speech samples. The rater listened to the word productions once only. The PCC was calculated as $100 \times (\text{number of correct consonants} / \text{number of correct and incorrect consonants})$ (Shriberg & Kwiatkowski, 1982). The ICC values for inter-rater and intra-rater reliability of PCC were 0.892 and 0.933 respectively in current research.

2.3.2. Kinematic assessment

During the kinematic task, each subject sat in a chair adjusted to 100% of their lower leg length. The trunk of the subject was secured to the chair-back with a harness to minimize trunk flexion and rotation. The facial model was modeled according to method described by Chen et al.¹⁶ Four 6-mm-diameter markers coated with infrared-reflecting material were attached to a facial mask at the forehead, bilateral pre-auricular areas and nose. These markers on mask help to establish a reliable coordinate system of head and minimize artificial error that might be caused by movement of facial skin (Fig. 1a). Additional five markers were attached to the bilateral corners of the mouth of the subjects, as well as to the central upper and lower lips and jaw (Fig. 1a).

All participants underwent mono-syllable (MS) and poly-syllable (PS) task assessments. The speech task comprised combination of a bilabial consonant (/p/) and five basic vowels (/a/, /i/, /u/, /æ/, and /o/) to form the target consonant-vowel syllable. We chose the bilabial consonants to elicit the lip opening-closing movement in each consonant-vowel syllable. For both tasks, the examiner pronounced the syllables themselves and asked participants to repeat

after the examiner. The examiner pronounced the target syllable(s) at a relatively slow rate for clarity purpose. During the MS tasks, participants were asked to randomly speak /pa/, /pi/, /pu/, /pæ/, and /po/ separately. During the PS task, participants were required to speak /pa, pi, pu, pæ, po/ in a sequence. In the MS task, each syllable was repeated ten times in a trial. In MS and PS, each task was repeated until ten available trials were completed. The examiner indicated the start of the task using a LED-light signal and vocal cue.

2.4. Data analysis

This study developed analysis coded using LabView (National Instruments, USA) language to integrate kinematic data and video images, and to process kinematic data. Reflective markers attached to the facial mask established a reference coordination system with a positive x, y, z orientation in the horizontal rightwards, anterior forwards and vertical upwards directions, respectively (Fig. 1b). The reference coordination system originated at the nose marker. Because most of the motion of the lip and jaw was confined to the vertical plane, the kinematic measures will focus on this single dimension (oral aperture in the z-axis). The overall utterance period of a speech task was determined based on the time interval between start of the first rising (opening) velocities and the end of the last decreasing (closing) velocity of the lower lip and jaw markers.

2.4.1. Spatial and temporal parameters

Peak vertical opening displacement of mouth (oral aperture) was determined for each speech task based on the maximum vertical distance between the upper- and lower-lip markers within the utterance duration. Peak vertical opening velocity was calculated by determining the maximum time derivatives of the vertical opening displacement. Furthermore, the CVs for utterance duration were obtained by dividing the standard deviation of utterance duration by the mean utterance duration. Larger CV indicates higher variability of utterance duration in speech tasks.

2.4.2. Articulatory coupling and synchrony

A cross-correlation analysis, as described in previous study (Green et al., 2000), was used to quantify the spatial (peak coefficient) and temporal coupling (lag) of lower lip and jaw during speech tasks.

Peak coefficients (r) and their associated time lags were derived from each cross-correlation functions computed between the treated displacement traces of lower lip×jaw articulatory pairs. Mean peak coefficients were calculated using Fisher's r -to- z transformation. The peak coefficient (r) is an index reflecting the relative similarity of the lower lip and jaw signals. The lag is an index of the synchrony of both signals. Low spatial and temporal coupling (low coefficients and high lags) reflect poor coordination between articulator pairs.

2.5. Statistical Analysis

Group differences in age, body height, body weight, PCC, utterance duration and CV of utterance duration, peak vertical opening displacement and velocity of the lip, peak vertical jaw velocity, and peak coefficient and lag were compared using an independent t-test. Furthermore, gender differences between groups were determined using a Chi-square test. The level of significance was set to $p < 0.05$.

3. Results

Demographic data did not differ significantly between SQ CP and healthy children (Table 1). The PCC ranged from 85.0-94.0% in children with SQ CP, and 92.0-100.0% in healthy children. Children with CP had significantly lower average PCC than did healthy children ($p < 0.001$) (Table 1).

Fig. 2 showed the averages of peak coefficient and lag values obtained at CP patients and healthy children for the lip lower \times jaw pair. Analytical results demonstrated that peak coefficients were not significantly different between SQ CP and healthy children in both tasks. But lag was greater between lower lip and jaw movement for SQ CP than healthy children in MS ($p=0.015$) and PS ($p=0.007$) tasks.

Kinematic results demonstrated that children with SQ CP displayed greater peak vertical opening jaw displacement in MS ($p=0.032$) and PS ($p=0.035$) tasks than healthy children (Fig 3). Children with CP also had greater peak vertical opening velocity of lower lip (MS, $p=0.024$; PS, $p=0.036$) and jaw movement (MS, $P=0.048$; PS, $P=0.02$) in both tasks than healthy children (Fig. 3). Furthermore, the CVs of utterance duration for children with SQ CP were at least twice of healthy children for both tasks (MS: 13.2 ± 6.8 vs 5.3 ± 4.0 , $p = 0.003$; PS: 15.2 ± 8.0 vs 6.5 ± 4.8 , $p = 0.010$). Nevertheless, the average utterance duration did not differ significantly between groups for both the MS and PS tasks (MS: 0.90 ± 0.17 (CP) vs 0.82 ± 0.07 (control), $p = 0.164$; PS: 0.92 ± 0.17 (CP) vs 0.84 ± 0.08 (control), $p = 0.113$).

4. Discussion

The present study is the first kinematic study to investigate the labiomandibular movement in children with SQ CP. In this study, we found that children with SQ CP displayed labiomandibular coupling movement impairment, especially the temporal coupling. Clinical assessment of motor speech can not comprehensively measure the underlying neuro-motor control for speech production, such as temporal-spatial coupling movement and oromotor variability. Using the current analysis, we observed that these children had high temporal oromotor variability with quantifying measures. With physical limitations, these children they need to make more efforts to control and coordinate the labiomandibular movement for speech production. The poor temporal and spatial coupling in CP children suggest their

inability to control muscle activations and de-activations and oromotor activities at varying degrees at the neuronal level. Thus, the approach used in the study is likely to provide a neuro-motor control model of the articulatory behaviors in this population. More importantly, the use of multiple measures in the current research offered an alternative to understanding the underlying abnormal labiomandibular control for speech production in CP.

It is interesting that children with SQ CP produced lower degree of temporal coupling, but not spatial coupling than healthy children in the labiomandibular movement. For speech production, each coordinated movement requires temporal and spatial control in the innervating muscles of the articulators, the larynx, and the chest wall (Smith, 2006). Successful temporal control depends on the appropriate timing control of muscle activations/de-activation and good spatial control refers to the appropriate graded activity control for speech production (Smith, 2006). It is likely that abnormal timing and sequence of muscle activity in children with CP is linked to the distortion of motor commands by transmission through damaged descending pathways (Maner, Smith, & Grayson, 2000). Generally, lower lip and jaw motion exhibited near-synchronous movement and well-formed movement trajectories for healthy children, consistent with previous research (Green et al., 2000). Oral opening involves temporarily moving the lips and jaw from rest or neutral position to a position, and changes in lip and jaw opening may results from direct modification of jaw and lip opening muscle activity (Gracco & Löfqvist, 1994). Green et al. (2000, 2002) suggest that the mandibular operating system assumes dominant responsibilities in early normal speech development. The very young children who exhibit early speech motor delays may have a negative prognosis if they also struggle with limited mandibular control (Green et al., 2000). People with articulation disorders are largely attributable to immature or interruptive mandibular activities (Dworkin et al., 2003). Thus, the deficits in temporal control in children with CP could lead to the immature or interruptive synchronous labiomandibular movements in speech production, which further cause articulatory problems.

It appears that children with spastic quadriplegia had high temporal oromotor variability for speech production. In this study, healthy children produced repetitions with relative regular intervals and low variations in syllable durations, whereas children with SQ CP exhibited increased CVs in syllable duration. Our previous study also found the children with mild CP had greater CVs of utterance duration than healthy children (Chen et al., 2010c). High variability on utterance durations in children with SQ CP might reflect deficits in relative temporal control for speech production. Temporal variability may be due to difficulty in maintaining constant speech duration and speaking rate during the speech task. The deficits in temporal control may arise from poor motor coordination (Smith, 2006; Smith & Zelaznik, 2004). The clinical implication is that the temporal stability of speech movement training may benefit for speech production in these children.

We also observed that children with SQ CP exhibited greater peak velocities and displacements at the lower lip and jaw for speech production than healthy children did, which suggests effortful speech production. The effortful speech production might be due to spastic or weak muscle. The neural system underlying jaw motor control for speech has been claimed to be more closely coupled with neural systems involved in vocalization (i.e., laryngeal and respiratory control), as compared to those of the lips and tongue (MacNeilage, 1998; McClean & Tasko, 2002). A child with weak tongue movement may use the jaw to compensate during speech (Yunusova, Weismer, Westbury, & Lindstrom, 2008). Another reason may be that a greater degree of jaw displacement is reflective of a poor postural control for maintaining a stable jaw opening (Maner et al., 2000), thereby driving a greater displacement and velocity of lips. These findings may suggest the abnormal peak velocity at the jaw may be used as an indicator of neurological impairment in children with SQ CP.

Children with SQ CP had impaired speech intelligibility. Results demonstrated lower PCC was found in children with SQ CP than in normal children. Even children with mild CP still had relatively lower speech intelligibility than normal children (Chen et al., 2010c). This study recruited CP subjects with speaking ability, as they achieve average PCC scores greater than 85%. Patients with spastic quadriplegia usually have more severely disrupted speech than speakers with spastic diplegia or hemiplegia (Chen et al., 2010b). To produce intelligible speech, the brain must generate motor commands to control activation of many different motor neuron pools that innervate the muscles for speech production (Smith, 2006). Damage of the immature brain in CP may cause variation in neural drive to muscles, resulting in higher speech variability and dysarthria, as a result, their speech intelligibility is reduced. The motor variability identified in the current research suggests that speech motor control with speech stability training is necessary to increase speech intelligibility in these children.

This study has some limitations, including the small number of participants, subjects' characteristics, and measurement methods used in kinematic analysis. We only enrolled children with SQ CP and relatively mild speech intelligibility impairment in the study. Therefore, our results can not be generalized to all cases of CP. The utterance durations are relatively long because the participants repeat the target syllable(s) at a relatively slow rate for clarity purpose. Despite these limitations, this study essentially provides clinician for a better understanding of the biomechanical and coordinative labiomandibular movement during speech production.

5. Conclusions

In this study, children with SQ CP displayed labiomandibular coupling movement impairment, especially the temporal coupling. The high CV values in utterance duration reflected children

with SQ CP had high temporal oromotor variability. High peak displacements and velocities of lip and jaw joint indicated children with SQ CP need more effort to control the labiomandibular movement for speech production. Information derived from this study may prove invaluable in the design of clinical treatment programs for these patients. Future studies may focus on kinematic assessment of different speech tasks and treatment strategies planning for children with CP of different severities.

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Figures Legends

Fig. 1. Experimental setup for kinematic analysis (a) reflective markers attached over the facial mask and oral areas; and (b) reference coordination system established based on mask marks.

Fig. 2. The illustration shows (a) vertical displacement trajectories for labiomandibular movement in a healthy child during mono-syllable task. Each signal has been centered about its mean; (b) the cross-correlation functions for signals lower lip and jaw. The peak coefficient and lag value were extracted from each cross-correlation function; (c) the average coefficients and (d) lags for lower lip and jaw in mono-syllable and poly-syllable tasks of children with quadriplegic CP and healthy children.

Fig. 3. The average peak opening (a) displacement and (b) velocity of lower lip and jaw movements in mono-syllable and poly-syllable tasks of children with quadriplegic CP and healthy children.