CHARACTERISTICS OF EPIGLOTTIC INVERSION IN CHILDREN WITH CEREBRAL PALSY

by

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B.A. Biological Sciences, Carnegie Mellon University, 2013

Submitted to the Graduate Faculty of

School of Health and Rehabilitation Sciences in partial fulfillment

of the requirements for the degree of

Master of Science, Speech Language Pathology

University of Pittsburgh

2016

UNIVERSITY OF PITTSBURGH

SCHOOL OF HEALTH AND REHABILITATION SCIENCES

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April 21, 2016

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This single cohort, retrospective, descriptive study, was conducted because of anecdotal, clinical observations that most children with cerebral palsy exhibit limited epiglottic inversion during swallowing. Puree bolus swallows from previously recorded Videofluoroscopic Swallowing Studies (VFSS) of children with cerebral palsy were analyzed with image processing software to compare the degree of the epiglottic angle before the swallow to the epiglottic angle at maximum rotation, and the competence of airway protection using the Penetration-Aspiration Scale. Data were analyzed to describe correlation between angle of epiglottic inversion and entry of material into the laryngeal vestibule. Descriptive statistics were used to describe means and standard deviation of epiglottic inversion, penetration-aspiration score, and the age and sex of the participants. A comparison was made between angle of epiglottic inversion in children with cerebral palsy and healthy norms. Findings of this study included significantly limited epiglottic inversion in children with cerebral palsy. A correlation was not demonstrated between epiglottic inversion and penetration aspiration scores.

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PREFACE

1.0   INTRODUCTION

Children with cerebral palsy (CP) are predisposed to dysphagia. The consequences of dysphagia include malnutrition, which can lead to delayed development and a higher incidence of infection (Kim, 2013). Few studies have characterized the kinematics of swallowing in this population. Efforts to more clearly understand swallowing kinematics in children with CP will ultimately benefit clinicians evaluating Videofluoroscopic Swallowing Studies (VFSS). Findings may guide clinical interpretation and information provided to families.

Cerebral Palsy: Cerebral palsy is a general diagnosis given to individuals who experience a nonprogressive motor disorder caused by damage in the brain of the fetus or infant. Cerebral palsy may affect sensation, perception, cognition, behavior, and communication (Schiariti, 2015). It is the most common physical disability occurring in early childhood. Cerebral palsy results from damage to the central nervous system (CNS). The damage can occur from in utero to 2 years of age with up to 80% of cases occurring in utero and 10% of cases occurring through asphxia during birth. After birth, cerebral palsy is most often caused by "central nervous system infection, trauma, strokes, and severe hypoxic events" (Erasmus, 2011). Cerebral palsy presents differently depending on the location and extent of the lesion. Though the lesion does not change with development, the presentation of the impairment may change depending on the ability of the CNS to adapt. Therefore, classifications are used to further differentiate presentations. These classifications are: spasticity, athetosis, rigidity, hypotonia, dystonia, or mixed. However, most

cases of cerebral palsy are classified by a non-specific code (Koman, 2004). Cerebral palsy is a highly variable and common disability.

Swallow Physiology: Swallow physiology can be described in five stages: oral preparatory, oral transit, stage transition, pharyngeal, and esophageal. During the oral preparatory stage salivation begins digestion, mastication breaks the bolus into an edible size and texture, and the bolus is positioned and contained. In the oral transit stage, propulsion of the bolus and velopharyngeal closure begin as the bolus leaves the oral cavity. During stage transition, hyolaryngeal excursion begins which starts the movement of the epiglottis and moves the airway out of the bolus path while applying traction force on the upper esophageal sphincter (UES). During this stage, the bolus enters the pharynx. In the pharyngeal stage, lingual propulsion and pharyngeal constriction push the bolus into the pharynx. Hyolaryngeal excursion continues to reposition the epiglottis in the horizontal position, the larynx is pulled toward the hyoid, and the UES is opened while the entry to the larynx is closed. The bolus and tongue muscles pull the epiglottis over the larynx, and finally, the bolus tail enters the UES (Coyle, 2014). Entry of the bolus into the UES begins the esophageal stage. Swallowing is a complex process. If any parts of this process are disturbed, dysphagia, or difficulty swallowing, may occur.

Dysphagia: Dysphagia is defined as a swallowing abnormality. Dysphagia can lead to "poor nutrition or dehydration, risk of aspiration which can lead to pneumonia and chronic lung disease, and less enjoyment of eating or drinking" (ASHA, 2016). Aspiration occurs as a result of the impaired interactions of many factors. These impaired interactions are caused by oropharyngeal dysphagia (OPD). OPD is a condition that disables normal transfer of the bolus safely from the mouth through the pharynx to the esophagus. OPD may be explained by many

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factors. Those specific to neurological damage (which is the cause of cerebral palsy) include damage to the cranial nerves in the brainstem swallowing center, cortical and subcortical areas of the brain, or motor neurons. Common complaints from patients with oropharyngeal dysphagia including, food "sticking", coughing and choking, weight loss, and lung infections (Rommel, 2015). Besides diet changes, OPD can be treated by a variety of compensatory strategies and exercises which may or may not be possible for a child with cerebral palsy depending on their neuromuscular or cognitive abilities.

Swallowing and Feeding Problems in Children with Cerebral Palsy: Swallowing and feeding disorders specific to children with cerebral palsy typically include delayed pharyngeal swallow initiation and reduced pharyngeal motility. Children with cerebral palsy also have a difference in "salivary parameters" which suggests an autonomic impairment (Ferreira, 2011). An autonomic impairment may affect digestion and ability to absorb nutrients. These difficulties with digestion are likely the cause of feeding aversions, which are also common in this population.

Dysphagia has previously been characterized in children with cerebral palsy. Kim et al. (2013) described findings from Videofluoroscopic Swallowing Studies (VFSS) on 29 children with cerebral palsy. In the severe cerebral palsy group, reduced lip closure, inadequate bolus formation, residue in the oral cavity, delayed triggering of pharyngeal swallow, reduced laryngeal elevation, coating on the pharyngeal wall, delayed pharyngeal transit, multiple swallows, and aspiration were significantly more common with increasing cerebral palsy severity. This study also showed that dysphagia is closely related to severity of cerebral palsy, with as many as 50% of children with severe cerebral palsy aspirating, mostly silently. The

authors therefore recommended early dysphagia evaluations for children with cerebral palsy, even without overt signs of aspiration. Though many characteristics of the swallow of children with cerebral palsy were investigated in this article, movement of the epiglottis was not characterized.

Mechanisms of Epiglottic Inversion: The epiglottis is a piece of cartilage at the base of the tongue which inverts over the airway during the swallow. It cannot invert on its own, but is moved as a result of a series of mechanisms which make up a healthy swallow. Mechanisms of epiglottic inversion were previously characterized, and therefore allow for a hypothesis to be made about its movement for children with cerebral palsy. Vandaele et al. (1995) determined which structures and mechanisms are responsible for epiglottic inversion. This mechanism was characterized through dissection of 20 adult cadavers and 5 VFS studies. The authors described how laryngeal elevation and anterior hyoid movement exerts traction to bring the epiglottis into a position below the horizontal. Two epiglottic movements were also characterized. In the first, the epiglottis is moved into a somewhat horizontal position by tongue movement. The second movement brings the epiglottis below the horizontal when the bolus passes through the pharynx, which is the result of biomechanical forces generated by movement of the hyoid bone and thyroid cartilage. Though this study described epiglottic inversion in adults, there are no available description of this mechanisms in children. When the epiglottis does not invert, it is indicative of impairments in other aspects of the swallow. A major contributor to epiglottic inversion is hyolaryngeal excursion, which is the central movement that protects the airway during the swallow. Because of the characteristics of swallowing in CP discussed by Kim et al. (2013), specifically delayed triggering of pharyngeal swallow, reduced bolus formation, and reduced laryngeal elevation, it is plausible that epiglottic inversion would be decreased in

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children with cerebral palsy. These characteristics are a reflection of both reduced tongue

movement and reduced HLE. Further, because limited epiglottic inversion is indicative of

impaired swallowing dynamics, the airway is likely more vulnerable to laryngeal penetration or

aspiration in this population.
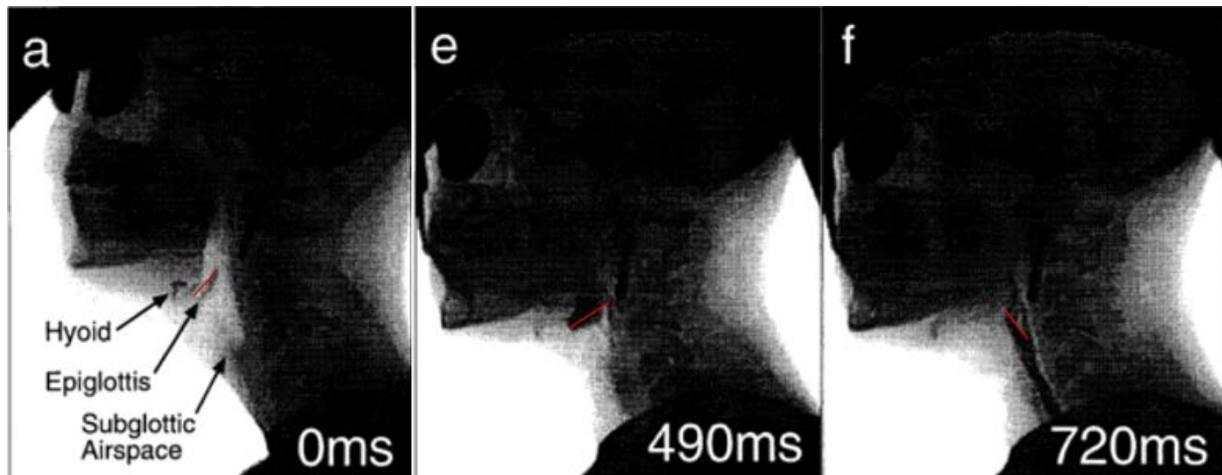
Figure 1: Mechanics of Epiglottic Inversion
In these frames taken from the Vandaele article, the epiglottis is at rest in frame "a". The first
epiglottic movement in seen in frame "e" in which tongue movement bring the epiglottis
into the somewhat horizontal position. The second epiglottic movement is seen in frame
"f" in which the epiglottis in brought below the horizontal by movement of the hyoid
bone and thyroid cartilage.

Kinematic Measures: In this study, kinematic measures were performed to measure total

rotation of epiglottic inversion. In order to measure epiglottic inversion, measures were aligned

to movement of spinal landmarks. Angles were calculated from the base of the vallecula to the

tip of the epiglottis. This allowed for an exact calculation of how far the epiglottis

rotated/inverted for each subject. This method is based on a previous study measuring hyoid

displacement. Steele et al. (2011) used an X,Y coordinate system in which the Y-axis was

aligned to with the spine (C2 to C4 vertebra) and the X-axis intersected perpendicular to the

spine. Structural movement tracing was used to calculate the displacement of the hyoid and

arytenoid cartilages relative to the spine. In the current study, the method also incorporated movement alignment with the spine and use of anatomical landmarks to measure movement. However, in the current study the anatomical landmarks are the base of the valleculae and the tip of the epiglottis rather than the arytenoid cartilages and hyoid.

To our knowledge, there has not been a previous study characterizing the epiglottic inversion in any children. Epiglottic inversion of aging adults has been characterized by Kang et al. (2010). In the youngest group for this study (≤45 years old) the smallest angle of epiglottic inversion was 89 degrees in a healthy adult. This number will therefore be used in the hypothesis as the smallest epiglottic angle which could occur in healthy individuals.

Clinically, limited epiglottic inversion of children with cerebral palsy has been anecdotally observed, however characterizing it in a systematic fashion will provide clinicians and researchers with information about the types of issues children with cerebral palsy may have with swallowing.

1.1   SPECIFIC AIMS AND HYPOTHESES

The aims of this research were to describe the epiglottic inversion of a small group of dysphagic children with cerebral palsy and to determine whether the degree of epiglottic inversion is predictive of swallowed material entering the airway (penetration-aspiration scale score) and the severity of airway compromise. It was hypothesized that epiglottic inversion would be limited to less than 89 degrees for most of the children with cerebral palsy. It was also hypothesized that

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fewer degrees of inversion would correlate with higher (worse) scores of the penetration

aspiration scale (PAS).

2.0   STUDY DESIGN AND METHODS

2.1   STUDY DESIGN

This is a descriptive, retrospective, single cohort study that made objective frame-by-frame measurements of epiglottic inversion during swallowing from previously completed videofluoroscopic studies. Epiglottic inversion data was compared to published values from prior research of epiglottic inversion. This comparision sample was obtained from the population most closely age-matched to the cohort of children with cerebral palsy. This study also included a correlational analysis between degree of epiglottic inversion and penetration-aspiration score.

2.2   RESEARCH METHODS

2.2.1 Ethical Considerations

Videos analyzed were collected in previous clinical VFS studies. Swallowing studies were completed with the patients in lateral view under videofluoroscopy and recorded on a Kay Pentax Swallowing Work Station. Studies were conducted using continuous fluoroscopy and a frame rate of 30 frames/second. Studies were performed by a speech language pathologist at a specialty rehabilitation hospital. These studies were taken before this study began for clinical purposes and were therefore not collected with a standardized approach. Investigators had no

control over methods used in these studies. Limitation of fluoroscopy time was ensured by the radiologist conducting each examination to account for the fact that children are more radiosensitive than adults. We obtained consents from the guardians of participants to use the pre-recorded videorecorded swallow study data for this study. Participants were also given the opportunity to provide assent, but all were unable to provide assent because of severe communication disorders. This study was approved by the Institutional Review Boards of both the University of Pittsburgh and the Children's Institute of Pittsburgh.

2.2.2 Inclusion Criteria

Eligible participants were children who (1) were diagnosed with cerebral palsy; (2) had swallowed at least one puree bolus during the videofluoroscopic examination; (3) were under the age of 18 at the time of the study. Signed consent forms from the guardians of the participants were also required for inclusion. Puree bolus was selected because it was presumed this consistency would be presented at similar volumes for each swallow, as puree is spoon fed. Puree consistencies in the VFSS included applesauce as well as general puree consistencies brought to the evaluation by the family of the participants.

2.2.3 Exclusion Criteria

Ineligible participants (1) were 18 or older at the time of the study; (2) did not swallow a puree bolus during the VFS study; (3) did not have a diagnosis of cerebral palsy. If consent forms from the guardians of the participants were not returned, these participants were not included.

2.2.4 Recruitment

Prior to IRB review, twenty-eight potential participants were identified who had both a diagnosis of cerebral palsy and a puree swallow during their VFSS. At this point, age was not a limiting factor in this study. However, following IRB review, it was determined that participants over the age of 18 would need separate consent forms sent to them if they were "decisionally able" or "not decisionally able". In our study, we did not have access to records which would allow us to determine this, so the participants were limited to less than 18 years old. Potential participants were then limited to a total of 18. Two of these participants were either deceased or moved without change of address provided so recruitment letters and consent forms were sent to 16 possible participants who underwent VFSS evaluations between the dates of 9/14/2007 and 8/28/2015. Recruitment letters were sent from the participants' speech-language pathologist, who had access to their addresses and medical records. The recruitment letter explained the study to the parents and also provided an explanation as to how consent or assent could be obtained. The consent form was written to the participants with the intention that it could be read to the participants by their parents if this was cognitively appropriate. An assent form was also attached which provided a space for the participant to sign if possible, circle a "yes" or "smiley face" if possible, or a space for the parent to describe why assent was not possible. A total of 5 letters were returned with consent to participate in this study.

2.2.5 Participants

Five eligible participants with cerebral palsy meeting all inclusion criteria were identified who had a VFSS between the dates of 9/5/2008 and 7/8/2014. These eligible participants consisted of 3 females, aged 3 and 10 years old and 2 males, aged 5 and 7 years old. A fifth eligible

participant (5 year old female) who provided consent was not included in the study because the quality of the video was not adequate for measurement. This was caused by constant head movements of the participant. Consequently, a total of 4 participants' VFS studies were analyzed (2 male, 2 female). Average age of participants in this pool was 6.25 years old with a standard deviation of 2.99 years. Participants were referred for evaluation for various reasons including swallowing difficulty, determining safety of eating, and poor weight gain. Studies were completed at a pediatric specialty rehabilitation hospital. Participant characteristics are described in Table 1. Diagnostic information such as etiology and type of cerebral palsy was not available.

Table 1: Characteristics of Study Participants

Participant	Age	Sex	Number of swallow studies	Number measurable swallows
1	5	M	1	1
2	10	F	1	1
3	3	F	2	10
4	7	M	1	4

2.2.6 Instrumentation

Kinematic analyses of all measures were performed using ImageJ (National Institutes of Health) image processing software on a Windows 7 personal computer.

2.3   DEPENDENT VARIABLES

The dependent variables were epiglottic rotation and PA scores.

2.3.1 Pre-study judgment training

Initial training: The PI was trained by the mentor who is an expert in the kinematic analysis of videofluoroscopic swallow data, including epiglottic rotation and penetration-aspiration scale

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scores. During training and before measuring any data from the present study, inter-rater reliability of PA scores was established using a set of VFSS videos used to train laboratory judges. Intra-rater reliability of epiglottic rotation measurements was also established using a set of VFSS studies used to train laboratory judges. Intraclass correlation coefficient (ICC) was computed to determine the degree of inter-rater reliability of PA scoring between the expert judge (the thesis advisor) and PI for PA scores using the ICC tool from the website of the Department of Obstetrics and Gynaecology of the Chinese University of Hong Kong. This tool calculates the ICC using the number of raters, number of cases, and a matrix for data. An ICC score of 0.872 was measured for PA scoring. Intra-rater reliability for measuring epiglottic rotation was also established by the principal investigator using non-study data. An ICC score of 0.9139 was measured. Following use of this tool, all data from both judges were entered into SPSS Version 23 to confirm the tool's accuracy. The results were identical, therefore the ICC tool was used for the remainder of reliability testing. Percent agreement within 1 PA point was used for PA scoring while percent agreement within 25 degrees was used for measuring epiglottic rotation (Tables 2 and 3).

2.4   MEASUREMENTS

All measurable puree swallows in each video were used. Both studies were measured for the patient who was evaluated by VFS studies on two separate dates. Because these studies were taken for clinical purposes, consistencies were not presented to each patient in a uniform manner. Puree was selected under the presumption it would be presented in a more consistent volume from study to study because it is spoon fed. Puree was also selected because it is a common

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consistency included in VFS studies. Every puree swallow for each participant was considered, however many could not be used because of movements of the participants or unclear anatomical landmarks. A total of 16 puree bolus swallows were analyzed for this study across all participants. Videos had been recorded at 30 frames per second.

2.4.1 Measurement of dependent variables: Epiglottic rotation

The video frame showing the epiglottis at rest before the onset of epiglottic inversion, and the video frame demonstrating maximal epiglottic inversion, were selected for plotting the epiglottic landmarks using ImageJ. The frame preceding the first rotational motion of the epiglottis that led to the pharyngeal swallow was selected as the "at rest" frame for each swallow, and the frame in which maximal epiglottic inversion/rotation had occurred was defined as the "maximal epiglottic inversion" frame.

        Four anatomic landmarks on each frame were identified (Figure 2): First, both the base of the vallecula and the tip of the epiglottis were plotted. Second, the anterior inferior corner of the C2 vertebra and the anterior inferior corner of the C4 vertebra were plotted. These two vertebral points formed a stationary participant-referenced spinal axis anchor, which was used to add or subtract gross rotational motion of the entire head that may artificially increase or decrease the measured epiglottic rotation (See figure 3 for example). ImageJ uses a Cartesian coordinate system to indicate degrees of rotation. The maximal angle of rotation was subtracted from the epiglottic angle "at rest" to determine the amount of epiglottic rotation. These two measurements were recorded both at rest and at maximum epiglottic inversion.

2.4.2 Measurement of dependent variables: Airway compromise

For each swallow, the 8-point Penetration Aspiration Scale (Rosenbek et al., 1996) was used to rate the depth of, and patient response to, laryngeal penetration and aspiration.
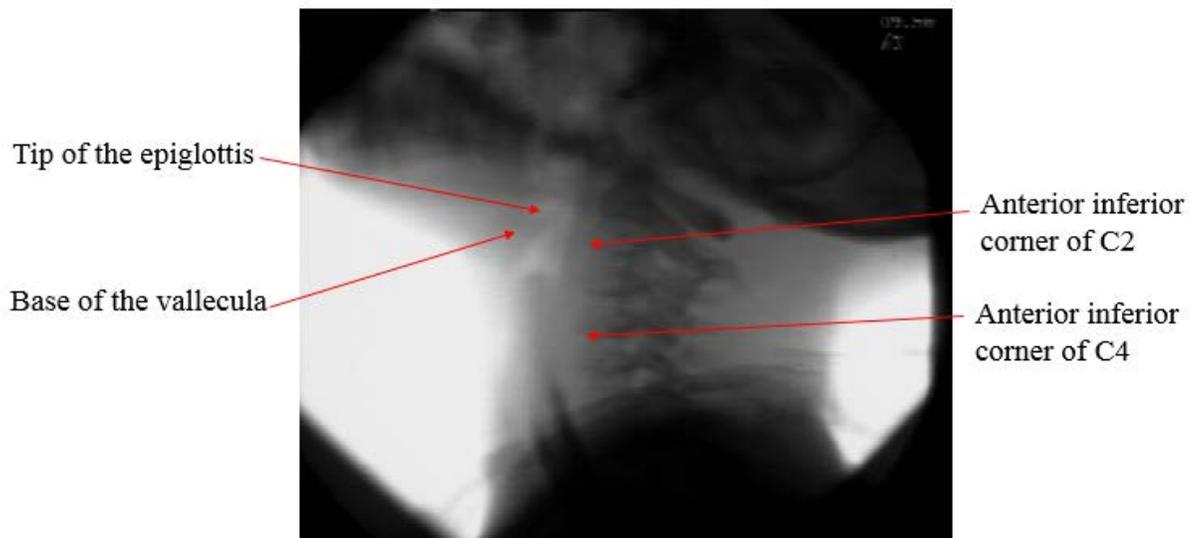

Figure 2: Landmarks Used to Measure Epiglottic Inversion

2.4.3 Data analysis

Epiglottic and spinal axis displacement measures from ImageJ were entered into a spreadsheet. Gross epiglottic rotation was calculated by subtracting the degrees of rotation present at maximal epiglottic rotation from the degrees of rotation present at rest. Head movement was then accounted for by subtracting (if clockwise) or adding (if counterclockwise) the degrees of spinal axis rotation from the degrees of epiglottic rotation to determine the angle of epiglottic rotation. See Figure 2 for example frames. In frames 1 and 3, the epiglottic angle is plotted. In frames 2 and 4, the spinal axis is plotted. Epiglottic rotation appears limited between frames 1 and 3, and spinal axis rotation between frames 2 and 4 increased apparent epiglottic rotation by rotating the entire head counterclockwise.

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2.4.4 Comparison to norms

The measurements from this study were compared to the only available norms for epiglottic inversion. Kang et al. (2010) investigated the influence of aging on movement of the hyoid bone and epiglottis in healthy adults. The youngest group in this study was less than 45 years old and the maximal angle of the epiglottis was measured using only liquid boluses. For data analysis, the average rotation of the epiglottis in the youngest group was used, 102.4 degrees. This average rotation was used for a one-sample t-test comparing this 1 average to all epiglottic inversion data from this study.

2.4.5 Establishment of judgment reliability.

Epiglottic rotation and PA score measurements were re-rated 2 times by both PI and the co-investigator (3 ratings total) for all swallows and the average of these measurements was used in the data analysis.
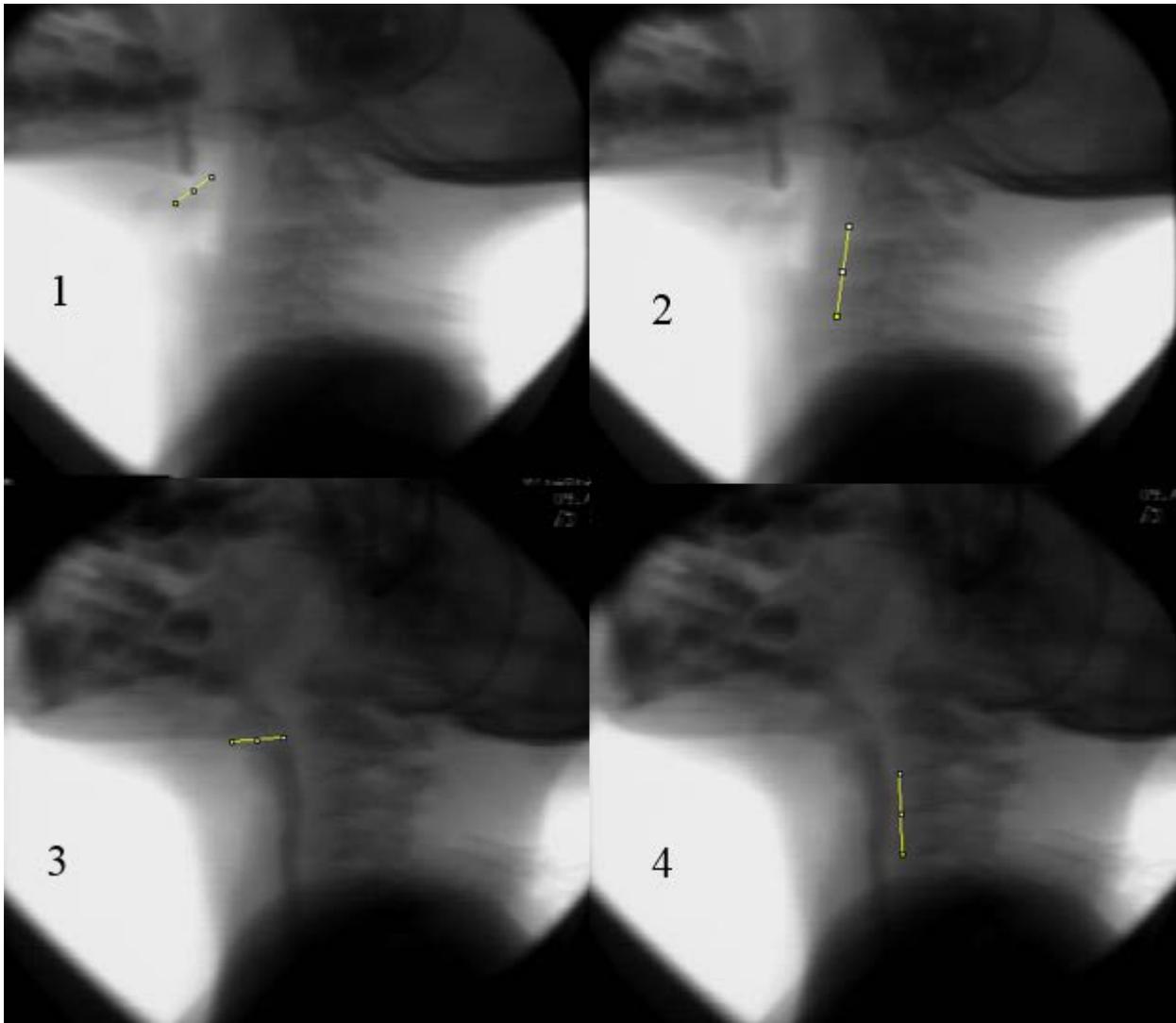
Figure 3: Measurement of Angles

The lines in each image correspond to the following: 1) angle of the epiglottis at rest 2) angle of the spinal y-axis at rest 3) angle of the epiglottis at maximum epiglottic inversion 4) angle of the spinal y-axis at maximum epiglottic inversion. The angle of the spine moved counterclockwise between images 2 and 4. This forward head movement was accounted for by adding the difference between the spinal angles in images 2 and 4 angle to epiglottic rotation values.

2.4.6 Measurement of Reliability:

Intra-rater reliability: The PI independently rated the dependent variable penetration-aspiration scores, and measured the two epiglottic dependent variables (epiglottic angle at rest, epiglottic angle at maximum rotation) for each swallow (Figure 2). This PI then randomly re-rated a randomly selected 50% of the analyzed swallows (8 swallows total) at least 2 weeks after initial measurement in order to determine intra-rater reliability. The intraclass correlation coefficient (ICC) determined test-retest reliability between the judge's first and second judgments for both dependent variables. ICC for intra-rater reliability of measurements of epiglottic inversion was calculated to be 0.6182. Fleiss (1986) defines ICC of between 0.40-0.75 "fair to good". This decrease from the ICC for training can be explained by the decreased definition for pediatric anatomy, and the movements of the participants. ICC for intra-rater reliability of the PA score was 0.7918.

Because each measure of epiglottic rotation used for the results is an average of 3 measurements, this allowed for two types of intra-rater ICC ratings. When determining correlation between the 3 measured rotations used to determine an average as the final measurement, the ICC result was 0.7875.

Inter-rater reliability: Inter-rater reliability was measured by having a second judge (a trained speech language pathologist) independently rate penetration-aspiration scores and epiglottic angles of the same randomly selected set of 50% of the analyzed swallows. ICC analysis for epiglottic inversion of 8 swallows was calculated at 0.5222. This fair to good correlation could be explained by a difference in training between the PI and second judge as well as the factors listed above.

The 95% confidence intervals seen for these measures show a wide range of confidence in the correlation. Because of this, there is a need for more intensive training in future studies. Further, the reliability measures were only tested for 8 swallows (50% of the total swallows) which decreases confidence.

Percent agreement: Percent agreement within 25 degrees was used to compare scoring for epiglottic rotation (Table 2). Percent agreement within 1 PA score value was used to compare PA scoring (Table 3). Lower percent agreement which could again be accounted for by differences in pediatric anatomy and movements of the participants.

Table 2: Summary of Percent Agreement - Epiglottic Rotation

Measurement Type	Percent Agreement within 25 degrees
Training: Percent agreement for epiglottic inversion in VFS studies of adults	100%
Study data: Percent agreement for epiglottic inversion measurement averages	100%
Study data: Percent agreement for epiglottic inversion between 3 judgements	88%

Table 3: Percent Exact Agreement - PA Scoring

Reliability Type	Percent Agreement within 1 point
Training: Inter-rater reliability for adult PA scoring	88%
Study data: Intra-rater reliability for PA scoring	62%
Study data: Inter-rater reliability for PA scoring	88%

Table 4: Intraclass Correlation Results – Epiglottic Rotation

Reliability Type	Intraclass Correlation Coefficient	95% Confidence Interval
Training: Intra-rater reliability for epiglottic inversion in VFS studies of adults	0.914	0.472-0.981
Study data: Intra-rater reliability for epiglottic inversion measurement averages	0.618	-0.72-0.911
Study data: Intra-rater reliability for 3 epiglottic inversion measurements used for averages	0.788	0.585-0.912
Study data: Inter-rater reliability for epiglottic inversion	0.686	-0.372-0.935

Table 5: Intraclass Correlation Results - PA Scoring

Reliability Type	Intraclass Correlation Coefficient	95% Confidence Interval
Training: Inter-rater reliability for adult PA scoring	0.872	0.755-0.915
Study data: Intra-rater reliability for PA scoring	0.792	0.296-0.954
Study data: Inter-rater reliability for PA scoring	-0.098	-7.180-0.794

2.5   STATISTICAL ANALYSIS

Descriptive data includes age and sex of participants (Table 1). Means and standard deviations of epiglottic rotation, PA score, age, and sex were calculated. Inferential statistics was deployed but were weakened by the small sample size and serial dependency of the data. The correlation between rotation and penetration-aspiration scores was determined using a Pearson correlation coefficient on SPSS Version 23 software and summarized visually using a scatterplot. A one-sample t-test was performed using SPSS Version 23 software to compare the mean rotation

values from this study to the mean from a study of normal adults (Kang, 2010). No reference

data for healthy children is currently available for comparison.

3.0   RESULTS

Table 6: Results of Epiglottic Inversion and Penetration Aspiration Score.

Swallow ID	Total Epiglottic Rotation (degrees)	PA Score
1_1	28.77	2
2_1	50.04	1
4_1	48.48	8
4_2	57.41	4
4_3	17.71	8
4_4	34.12	4
4_5	14.84	1
5_1	47.99	1
5_2	72.93	1
5_3	55.62	1
5_4	70.53	8
5_5	53.83	1
6_1	42.36	1
6_2	2.04	1
6_3	17.32	1
6_4	9.06	1

The results of the VFS studies showed that epiglottic rotation in children with cerebral palsy is limited. Consistent with the hypothesis that epiglottic inversion would be limited to less than 89 degrees, the highest angle of epiglottic inversion recorded was 72.93, 16.07 degrees less than the lowest rotation in a group of healthy adults under 45 years old. Forty-four percent (7/16) of the measurable swallows exhibited less than 35 degrees of rotation. The average rotation for children with cerebral palsy was 38.94 degrees, significantly less than the normal adult rotation average of 102.4 degrees. The standard deviation was higher for children with cerebral palsy at 21.9 degrees while the standard deviation was 13.4 degrees for normal adults under 45 years old. However, this may be artifact of our small sample size. The one-sample t-test showed epiglottic rotation in children with cerebral palsy was statistically significantly lower than the normal adult population (<45 years old), $t(15) = -11.011$, $p = .000$.

Average PA score was 2.75 with a standard deviation of 2.79. However, visual inspection of the plotted PA scores and epiglottic rotation angles (Figure 3) did not demonstrate any correlation between the two, which does not support our hypothesis. In fact, there is a slight increase in penetration aspiration score with increased epiglottic inversion (Figure 4) though this may have been due to the small amount of data points. This slight correlation is also seen following removal of outliers (Figure 5). A Pearson correlation also showed no significant correlation ($R=0.186$, $p =0.492$). Aspiration occurred with epiglottic inversion angles of 17.71, 48.48, and 70.53 degrees. From these data, it cannot be concluded that there is a correlation between epiglottic inversion and penetration aspiration scores for children with cerebral palsy.
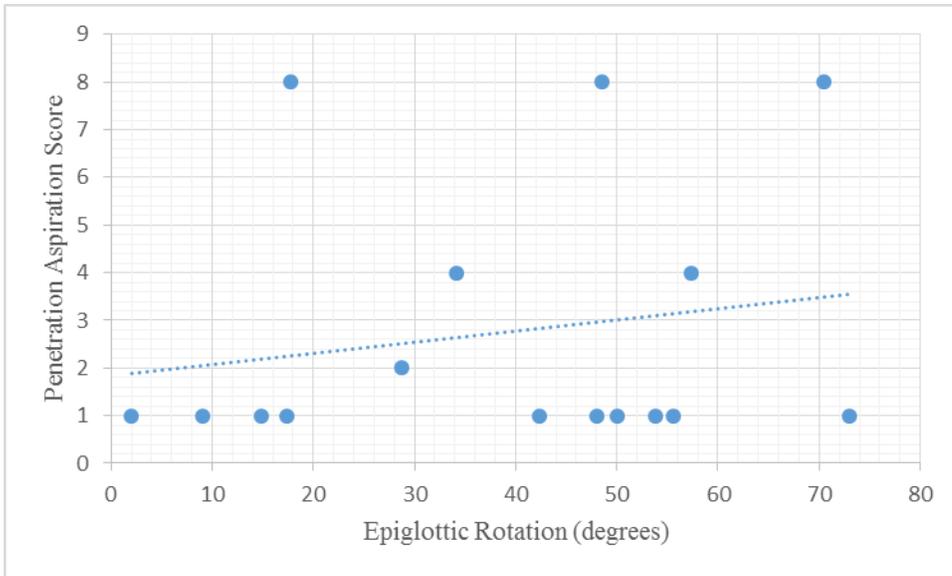
**Figure 4: Relationship between Epiglottic Inversion and PA Score (p>.05)**
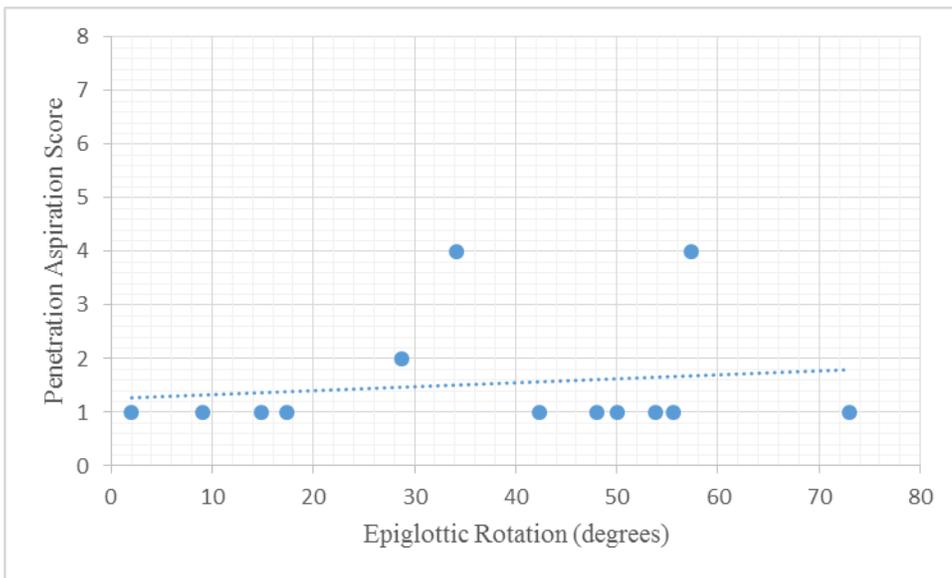
Figure 5: Relationship between Epiglottic Inversion and PA score without outliers (p>.05)

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4.0   DISCUSSION

This study investigates a new method of analyzing the kinematics of swallowing. In this study, rotation of the epiglottis was measured in order to obtain more information about swallowing kinematics in an at-risk population. It is a new method which compensates for head movement. Head movement is a symptom of motor disorders which may may occur in populations beyond those with cerebral palsy, such as patients with Parkinson's Disease. We were able to obtain data from these novel measures with fair to high reliability for most measures.

The results of our study indicated that epiglottic inversion during swallowing in children with cerebral palsy is less than that seen in healthy young adults. Although this may be an artifact of differences in aerodigestive tract anatomy between children and adults, this preliminary study provides the first measurement of epiglottic rotation of persons in this population, and useful data regarding swallow kinematics in children with cerebral palsy who are disproportionately affected by dysphagia. Given that research into swallowing physiology using x-ray imaging in healthy human children is considered ethically undesirable, our comparison to young adults represents the only plausible comparison available. Studies comparing these same dependent variables between groups of children with cerebral palsy and may shed more light on this question.

The hypothesis that epiglottic inversion would be limited for children with cerebral palsy was supported by our investigation of this small sample, as all epiglottic inversion was less than

89 degrees. Nearly half of the swallows analyzed failed to produce more than 35 degrees rotation. The limited epiglottic inversion for children with cerebral palsy is likely explained by the decreased HLE and tongue movement previously studied in this population. Because the epiglottis is a piece of cartilage, it is moved by other muscles in the swallowing mechanism such as tongue muscles and the muscles involved in HLE. If HLE or tongue movement do not occur, the epiglottis will not be inverted and will not contribute to closure of the supraglottic passage. Mechanisms of spasticity and athetosis occurring in cerebral palsy likely explain this change in swallowing kinematics. However, because of our small sample, they warrant further investigation with stratification of participants by type of motor impairment.

The hypothesis that fewer degrees of inversion would correlate with higher (worse) scores of the PAS was not found to be true in this study. Surprisingly, there was not a strong correlation between reduced epiglottic inversion and airway penetration and aspiration. This is counterintuitive because complete epiglottic inversion is a component of airway closure. However, this result may have been caused by a small sample size that produced a wide range of PA scores over a small number of swallows, and predominance of swallows in our data set from one participant. Naturally, a larger sample would have provided a more valid sampling of this population's swallow kinematics. Another explanation as to why this hypothesis was not supported is that closure occurs from the epiglottic petiole to the arytenoids, as opposed to epiglottic inversion for protection (Logeman, 1992). This may represent a compensatory adaptation that ensures airway protection, which is necessary for physiologic survival. Other explanations may include that epiglottic inversion is not essential for airway protection (Medda, 2002), or that in children, anatomical orientation of structures lowers airway protection dependency on epiglottis coverage of the laryngeal vestibule.

Research has shown that it is common for children with cerebral palsy to experience swallowing problems (Kim, 2013). This study suggests that limited epiglottic inversion is included as a swallowing characteristic for children with cerebral palsy. Limited epiglottic inversion may contribute to increased residue in the valleculae which may further put the participant at risk for aspiration. It is likely the limited inversion is caused by irregular movement and posture development. Because the epiglottis is inverted by the movement of muscles, it can be inferred that children with cerebral palsy have limited contraction strength (weakness) or efficiency of contraction (due to antagonist muscle resistance) of the following suprahyoid muscles: digastric, geniohyoid, and mylohyoid muscles (Figure 6). These muscles "appear to be the primary effectors of anterior hyoid bone movement, and thus are the principal muscles affecting epiglottic movement" (Vandaele, 1994). Underlying motor issues are likely the cause of limited epiglottic motility.
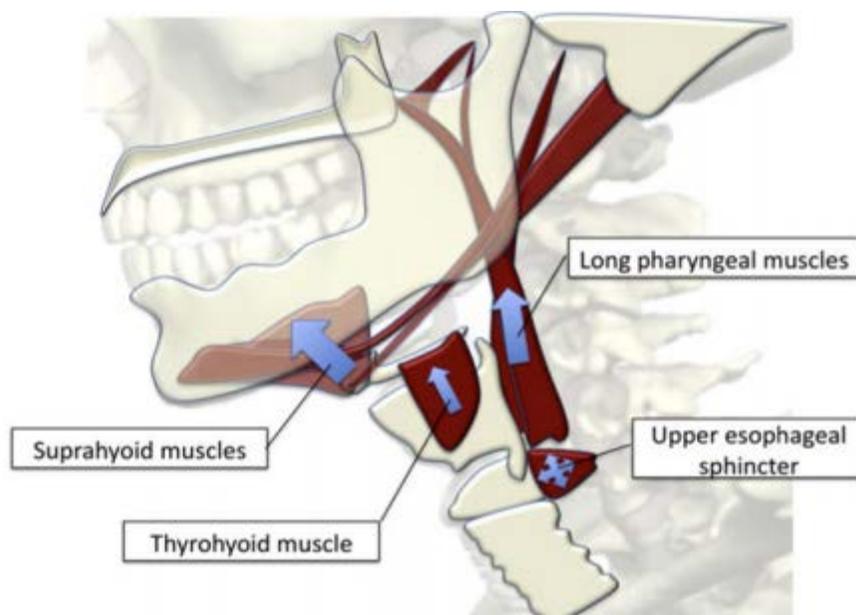

Figure 6: Suprahyoid muscles
(Source: Pearson, 2013) The digastric, geniohyoid, and mylohyoid muscles are all suprahyoid muscles. Contraction of these muscles causes anterior hyolaryngeal excursion and subsequently epiglottic inversion.

4.1 LIMITATIONS AND SUGGESTIONS FOR FUTURE RESEARCH

There were several limitations in our study's design. The small sample size decreases generalizability. In terms of reliability, there is a strong need for increased training in future studies, as demonstrated by the wide range of confidence intervals. Also, the type and severity of cerebral palsy were unknown to the principal investigator, who only had access to the VFSS, age, and sex of each participant. This additional information would have been useful because cerebral palsy is such a varied disorder. By noting the subtype and severity, as well as the predominating form of motor impairment, more useful conclusions might have been drawn which could point to both the etiology and clinical significance for this diverse population. Children with cerebral palsy have also demonstrated variability in mobility across settings (Tieman, 2007). This variability could account for the differences seen between the 2 dates of VFS studies of one participant. For future research, variability of swallowing at different days and times may be of interest.

It would also have been helpful to have knowledge of the norms for epiglottic movement in typical children instead of adults. However as discussed earlier, there are ethical concerns about exposing healthy children to x-rays for research purposes. Knowledge of norms for the pediatric population would have helped draw appropriate conclusions about the extent of the limited inversion. Without this data, it is not possible to be confident that limited epiglottic inversion is not seen in the general population in pediatrics.

We also did not know the exact bolus volumes that were administered by clinicians during the VFS studies. The epiglottis is partially inverted by the size of the bolus. If the bolus is not consistent it cannot be clearly determined that an increase or decrease in epiglottic inversion

is not caused by a difference in bolus size. For future research, a consistent bolus volume would remove that possibility.

Further, comparison data for norms should also include equivalent bolus consistencies. The adult epiglottic rotation was measured with a liquid bolus while this study measured with a puree bolus. As mentioned above, epiglottic inversion is affected by the type and size of the bolus. In future studies, comparisons should be made across the same bolus consistency and size.

Lastly, the number of measureable swallows for each participant should be consistent. In this study, the majority of measured swallows were from one participant. This caused a skew in the data that decreased generalizability. By analyzing an equal number of measurable swallows, the confidence in conclusions would increase.

For future research, more control of variables could be obtained by a prospective rather than retrospective study. The volume of the bolus should be controlled as well as the number of measurable swallows per participants. This consistency in measurements would increase confidence that the measured difference in epiglottic inversion are caused by the motor impairment instead of other factors such as skew in the data, or bolus consistency or volume.

4.2   CLINICAL IMPLICATIONS

The results shed light on some of the possible biomechanical explanations for misdirection of swallowed material during swallowing in children with cerebral palsy, who have elevated prevalence of pneumonia. Aspiration while swallowing can be a major contributor to pneumonia pathogenesis. Because aspiration is prevalent in this population, careful evaluation of the aerodigestive kinematics rather than global manifestations of cerebral palsy are warranted.

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The results of this study increase awareness into the motoric and swallowing related impairments of children with cerebral palsy. This information can be shared with clinicians, patients, and their families and discussed when interpreting VFS studies. Through motion analysis, researchers are able to begin to differentiate neuronal damage and hypothesize treatment strategies. Limited epiglottic inversion indicates underlying muscle impairment. In this population, these findings are important because they increase the awareness of the types of issues children with cerebral palsy may have with swallowing and may lead to future research to lower the risk this impairment poses.

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