

Physiological Variability in the Deglutition Literature: Hyoid and Laryngeal Kinematics

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Abstract A literature review was conducted on hyoid and/or laryngeal displacement during swallowing in healthy populations according to several inclusion criteria. Anterior and superior displacement measures of both structures from previously published studies were compiled for meta-analysis. Results showed a large degree of variability across studies for each structure and plane of movement. Potential sources of variation were identified, including statistical, methodological, stimulus-related, and participant-related sources.

Keywords Swallowing · Deglutition · Hyoid · Larynx · Kinematics · Movement · Deglutition disorders

Swallowing disorders (also known as dysphagia) are common following neurological impairments (such as stroke and brain injury). Dysphagia is also a major concern following head and neck procedures and in progressive illnesses (such as Parkinson's disease). Dysphagia can significantly impact a person's quality of life as well as their health status. Individuals with dysphagia are at risk for aspiration (material passing below the vocal folds and entering the lungs) and for developing pneumonia.

The gold-standard tool for assessment of dysphagia is the videofluoroscopic swallowing study (VFSS) [1]. It is a radiographic imaging protocol in which various foods and

liquids are mixed with a contrast medium and swallowed under fluoroscopy allowing for direct visualization of swallowing physiology. The VFSS allows clinicians to assess the safety and efficiency of swallowing across various textures and the impact of compensatory maneuvers on swallowing function. Also, measures of structural displacement and temporal intervals can be made during VFSS analysis. Unfortunately, the VFSS procedure must be limited to a brief number of swallows (usually between 3 and 10) [2] to limit the patient's exposure to radiation. The brevity of the exam makes interpretation susceptible to variable performance; this challenges clinical decision-making and scientific analysis.

Swallowing physiology can be quantified both spatially [3–6] and temporally [7–12]. Most prior studies of swallowing physiology in the dysphagia literature report measures of data dispersion (e.g., standard deviations); however, very few studies acknowledge or address the sizable variability in their data. Variability in swallowing physiology may threaten the representativeness and reliability of assessments and obscure change post-treatment. In this article we describe, synthesize, and discuss the physiological variability of hyoid and laryngeal displacement that is reported in the swallowing physiology literature.

Significance of Variability

From a clinical perspective, life-altering decisions regarding safety for oral feeding and/or compensatory strategies are based on a brief set of swallows. How confident can the clinician be that this “snapshot” view is representative of the patient's typical performance? Management and treatment decisions may be overly cautious or dangerously bold in the presence of variability on the VFSS. From a

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scientific perspective, how does variability threaten data representativeness if the presentation captured by the “gold standard” is itself variable? Furthermore, in scientific studies of treatment efficacy, what is the impact of such inherent variability on the ability to measure and detect meaningful outcomes?

Certainly, some authors have acknowledged that intra-subject and intersubject variability in swallowing physiology may play an important role in the interpretation of dysphagia research. For example, Kendall et al. [13] suggest that “intrasubject variability should be analyzed and will provide information about changes in swallow sequences in the same subject at the same sitting and at different sittings.” Lof and Robbins [11] documented the existence of “high variability from trial to trial in some normal subjects” on several duration measures and suggested that “individual performance must be carefully considered when one is evaluating the potential influence of treatment.” Despite this, a systematic investigation into the variability of swallowing on VFSS has not yet been conducted. The meta-analysis in this article takes a first step in this direction by synthesizing the results of the published literature on anterior and superior displacement of the hyoid and larynx during swallowing. These movement trajectories are known to facilitate airway closure [14] and upper esophageal sphincter opening [15].

Methods

Search Strategy and Inclusion Criteria

A literature search was conducted in June 2010 to find reports of hyoid and/or laryngeal displacement in healthy populations. The electronic databases PubMed and MEDLINE were used with a combination of the following search terms: *swallowing*, *deglutition*, *dysphagia*, *hyoid*, *larynx**, *hyolaryngeal*, *displacement*, *kinematics*, and *movement*. The search was limited to studies published on healthy adult humans in English. After removing duplicates, the search yielded 164 publications, which were reviewed for the following inclusion criteria: (1) VFSS as the imaging tool; (2) intact head and neck anatomical system; (3) displacement data of hyoid and/or larynx along with standard deviation (SD) or standard error of the mean (SEM); (4) reporting of both anterior and superior displacement on regular-effort liquid swallows; (5) Cartesian coordinates rotated to an anatomical plane of reference (e.g., spine); and (6) use of a scalar reference to control for magnification. After applying these criteria, 13 studies remained for in-depth analysis. The reference lists of these publications were hand-searched for additional articles but yielded no additional articles meeting the inclusion criteria.

Studies Included

A brief overview of each study that met the inclusion criteria is provided below. We restrict this overview to a discussion of methodology related to hyoid and laryngeal displacement with thin-liquid swallowing, although most of these studies involved several other measures and stimuli.

Dodds et al. [3] examined hyoid displacement of 15 (9 male) participants (age = 39 ± 19 years) who were referred for upper GI assessments but had no history or evidence of oropharyngeal dysphagia. Each participant swallowed two boluses of 2, 5, 10, 15, and 20 ml of 250% weight/volume (w/v) barium. Hyoid displacement was measured by taking the difference between the hyoid positions at rest and at maximum displacement. Data were presented as an average group value at each bolus volume and data dispersion was reported using SD.

Dantas et al. [16] reported hyoid and laryngeal displacement data from 10 healthy male volunteers who swallowed two boluses of each volume (2, 5, 10, and 20 ml) of 40% w/v barium. Hyoid displacement was calculated by comparing the rest frame to the peak frame. Data were presented as average group value at each bolus volume. Dispersion was reported in the form of SD.

Van Daele et al. [17] reported data for hyoid bone displacement from 5 males (3 in their 20 s, 2 in their 70 s). These were individuals who were referred for VFSS but judged to have normal swallowing. Each participant swallowed two 5-ml boluses. The results were averaged for each participant and dispersion was reported as SEM.

Perlman et al. [18] reported hyoid displacement data for 20 male participants (ages unknown) in two groups: half with “normal” hyoid and half with “reduced” hyoid (judged subjectively). Analysis was based on comparison of the hyoid position at rest and maximum. Each participant swallowed two boluses of an uncontrolled volume (from a cup containing either 5 or 10 ml), and each swallow was treated as distinct (not averaged) within the analysis. Only the results for the “normal” hyoid group are included in this meta-analysis.

Logemann et al. conducted two studies using identical methodology. One reports data on 16 healthy men [19] and the other on 16 healthy women [20]. In each study, the 16 participants are evenly split into young (20 s) and old (80+) and were required to swallow two 1-ml and two 10-ml boluses of “watery” barium each. Analysis was conducted using a frame-by-frame method to determine the hyoid and larynx movement trajectories. Grouped data are reported both by age and by volume. Each swallow was kept distinct in the analyses and dispersion was reported as SEM.

Ishida et al. [21] provided hyoid displacement from 12 healthy volunteers (7 male) aged 20-28 years. Each participant swallowed one 10-ml bolus of 50% w/v barium. Hyoid data were extracted by analyzing the swallow in a frame-by-frame fashion. Data were grouped by sex and dispersion was reported using SD.

Kim and McCullough [5] report hyoid displacement data from 40 (20 male) healthy volunteers in two age groups (20-51 and 70-87 years). The participants each swallowed four boluses of 50/50 barium sulfate and water: two at 5 ml and two at 10 ml. Hyoid displacement was extracted by comparing a rest frame to a maximum frame and dispersion was reported in the form of SD. While this study lacked a known scalar, the length of the C3 vertebra was assigned a fixed value of 15 mm to control for magnification. The swallows for each participant were averaged at each volume. Data were presented for each age group at each volume.

Paik et al. [22] provided data on hyoid movement trajectories in both healthy individuals ($n = 9$) and individuals with dysphagia (stroke, $n = 7$; myopathy, $n = 3$). The mean age of each group was in the early 60s. Each participant swallowed one 5-ml bolus of 35% w/v barium solution. Hyoid trajectories were tracked using a frame-by-frame method. Data were reported by group (control, stroke, myopathy), with SEM as the measure of dispersion. Only data from the healthy controls are included in this meta-analysis.

Kim and Han [23] reported raw hyoid displacement data for 12 young healthy male volunteers (age = 28.6 ± 3.0 years). Each participant swallowed one 5-ml bolus of 35% w/v barium. Hyoid displacement was extracted via a frame-by-frame analysis. Means and SD were calculated from the raw data for comparison in this meta-analysis.

Kang et al. [24] examined the hyoid displacement trajectories of 69 healthy volunteers (20 male) ranging in age from 26 to 78 years old. Hyoid displacement was stratified into four age groups: <45 ($n = 16$), 45-55 ($n = 21$), 55-65 ($n = 16$), and >65 ($n = 16$). The distribution of sex across the age groups was not reported. Each participant swallowed one 2-ml bolus of 35% w/v barium. Hyoid displacement was extracted using a frame-by-frame method. Data are reported by age group with SD as the measure of data dispersion.

Kim et al. [25] compared hyoid and laryngeal movement trajectories in 8 healthy controls (4 male, mean age = 64.6 ± 11.2 years) with those of 8 pneumonectomy patients (4 male, mean age = 65.4 ± 13.2 years). Each participant swallowed one 5-ml bolus of 35% w/v barium. Movement trajectories of the hyoid and larynx were extracted in a frame-by-frame fashion. Raw data (individual scores) were reported allowing for calculation of mean and standard deviation. The data from the dysphagia group are not analyzed in this article.

Bingjie et al. [26] reported hyoid and laryngeal displacement data for 105 stroke patients (57 male, mean age = 65.2 ± 8.2 years) with dysphagia and 100 healthy male controls (mean age = 62 ± 9 years). Patients with dysphagia were split into aspirators and nonaspirators in the analysis. Each subject swallowed one 3 ml and one 10 ml bolus of thin liquid barium. Movement trajectories were reported for each group (control, nonaspirators, and aspirators) at each volume (3 and 10 ml). Only data from the healthy controls are included in this meta-analysis.

Data Extraction

The mean values of both anterior and superior displacement of the hyoid and/or larynx were extracted from these publications for meta-analysis. Data extraction was limited by those variables that were reported in the original publications, given that the primary factors of interest varied across publications (i.e., group, age, bolus volume, sex). All measures were converted to millimeters for uniformity.

The corresponding measure of dispersion, SD or SEM, was also extracted from each publication. Two studies [23, 25] reported raw data, which allowed us to calculate the mean and SD manually. It is important to recognize that the SEM and SD are different mathematical functions. The SD is a true measure of data dispersion because it demonstrates how tightly each data point clusters around the mean [27]. The SD is calculated by squaring each observation, subtracting this from the mean, summing all the results, dividing this result by the total number of observations, and finally taking the square root of this value. The SEM is, by definition, always smaller than the SD because it is calculated by dividing the SD by the square root of the number of observations tested. The SEM is the standard deviation of the sampling distribution and demonstrates how representative the sample mean is of the true population mean [28]. The appropriate measure of dispersion for observational kinematic studies is the SD.

Next, confidence intervals for each study/variable were calculated. This was achieved by multiplying a specific t value (two-tailed, $\alpha = 0.05$, at $n - 1$ df) by the SD/[SQRT(n)]. The product of these, plus and minus the mean, gives the 95% confidence interval for that specific mean. These findings were plotted on a modified forest plot for each structure (hyoid and larynx) in each direction (anterior and superior) and can be found in Figs. 1, 2, 3, and 4. On all figures the circles represent the mean (in mm) for each particular study (or variable, where applicable) and the error bars represent the spread of the 95% confidence interval. The study author appears to the left of the mean, with the corresponding variables (if applicable) on the right. The scales for each dimension (anterior and superior)

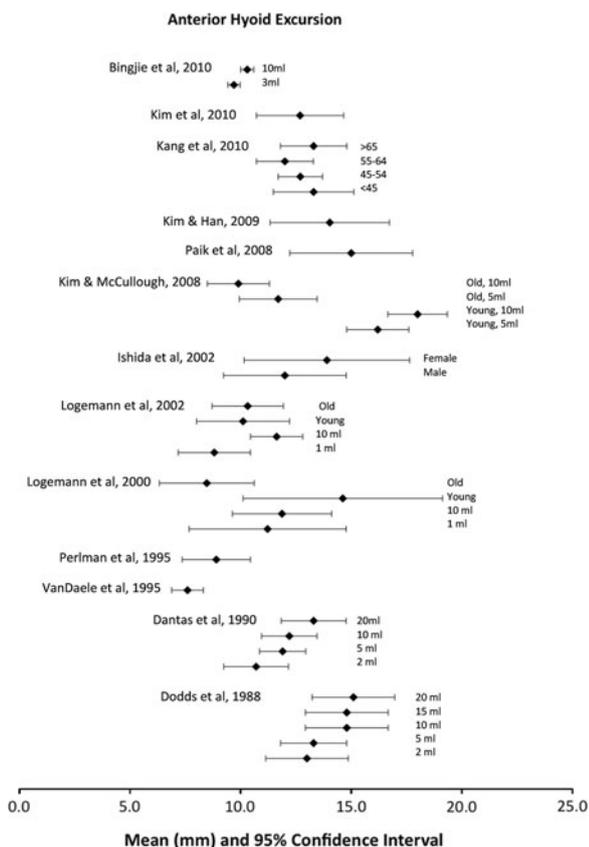


Fig. 1 Mean and 95% confidence intervals for anterior hyoid excursion from the 13 studies that met inclusion criteria. *ml* milliliters, *mm* millimeters

are held constant to allow for transparent comparison across structures (hyoid versus larynx).

Results

Hyoid Displacement

Figure 1 displays the results for anterior hyoid displacement in healthy adults. The mean anterior displacement ranges from 7.6 mm in the Van Daele et al. study [17] to 18.0 mm in the Kim and McCullough study [5]. First, it is apparent that there is a wide degree of variability across these 13 studies despite the strict inclusion criteria applied to the selection of articles for this meta-analysis. Each study that includes a factor of bolus volume [3, 5, 16, 19, 20, 26] shows a consistent increase in mean anterior hyoid displacement with increasing bolus size (with the exception of the elderly group in the Kim and McCullough study). However, the mean displacement magnitude seen for a particular volume (e.g., 10 ml) varies greatly across studies. For example, the mean anterior displacement value

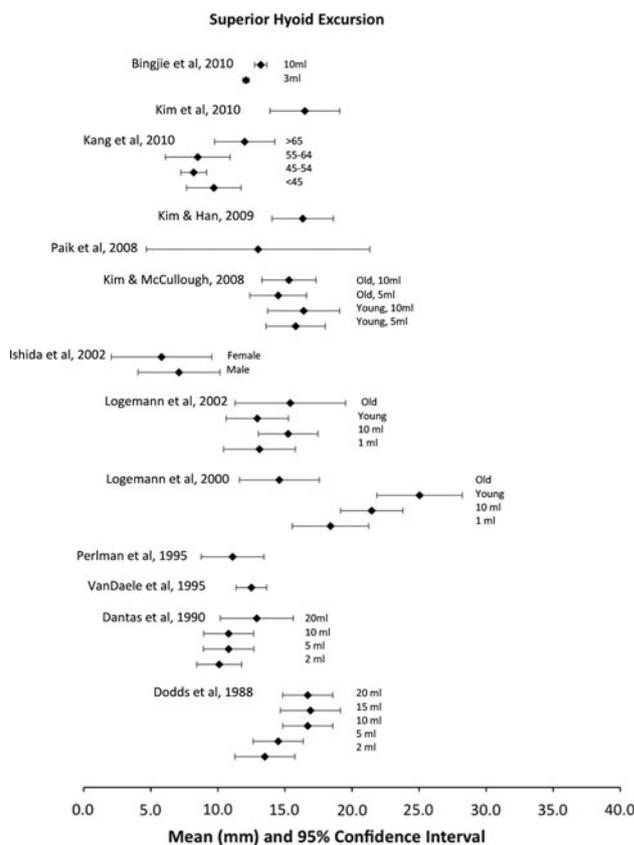


Fig. 2 Mean and 95% confidence intervals for superior hyoid excursion from the 13 studies that met inclusion criteria. *ml* milliliters, *mm* millimeters

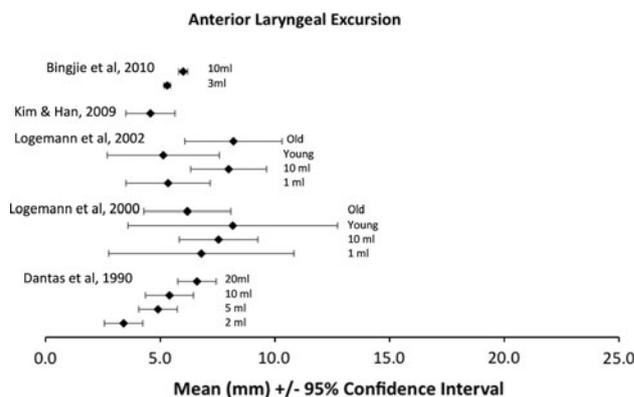


Fig. 3 Mean and 95% confidence intervals for anterior laryngeal excursion from the five studies that met inclusion criteria. *ml* milliliters, *mm* millimeters

for 10-ml swallows in the Dodds study [3] was 14.8 mm (CI: 12.93-16.67), in the Dantas study [16] it was 12.2 mm (CI: 10.94-13.96), in the young group in the Kim and McCullough study [5] it was 18.0 mm (CI: 16.66-19.34), while for the older group in the same study it was 9.9 mm (CI: 8.39-11.31). Thus, a substantial degree of variability is

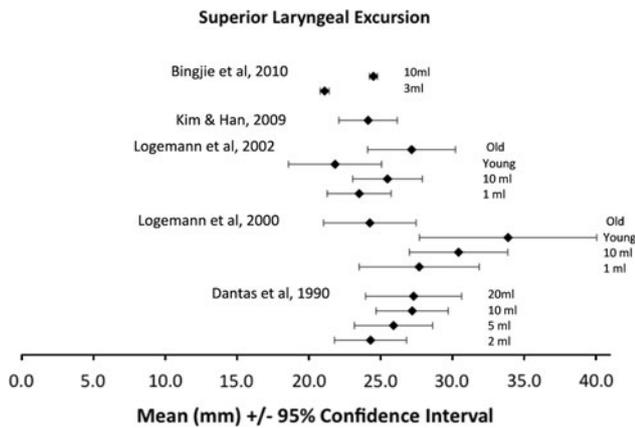


Fig. 4 Mean and 95% confidence intervals for superior laryngeal excursion from the five studies that met inclusion criteria. *ml* milliliters, *mm* millimeters

present across studies measuring anterior hyoid displacement. It is a well-known statistical fact that variability tends to decrease with increases in sample size. With the exception of Bingjie et al. [26], all the mean values shown in Fig. 1 are taken from sample sizes of less than 40, with many of them including fewer than 10 data points for each particular variable. The study with the tightest confidence intervals was also the study with the largest sample size [26]; this pattern will be seen across all four figures (anterior hyoid, superior hyoid, anterior larynx, and superior larynx).

The corresponding data for superior hyoid displacement are presented in Fig. 2. In this case, the range of variability for superior displacement is even greater than that seen for anterior displacement. The smallest mean superior hyoid displacement comes from the Ishida et al. study [21] at 5.8 mm, while the largest is 25.0 mm from the Logemann study in men [19]. Once again, there appears to be a consistent pattern of increasing displacement with increasing bolus size within studies but quite different values are seen for a particular bolus size across studies.

Laryngeal Displacement

Anterior laryngeal displacement is shown in Fig. 3. The smallest mean measure of anterior laryngeal displacement in the literature was 3.4 mm [16] while the largest was 8.2 mm [19, 20]. There appears to be less variability in anterior laryngeal displacement across studies compared with anterior hyoid displacement (Fig. 1). Once again, the trend of increased displacement for larger bolus volumes is seen within studies but with a wide range of values across studies.

Figure 4 shows the compiled data for superior laryngeal displacement, which ranges from a mean of 21.1 mm [26] to 33.9 mm [19]. The displacement values and confidence

intervals span a relatively large range across studies. Again, we see highly variable outcomes for a particular bolus volume (e.g., 10 ml) across studies.

Discussion: Potential Sources of Variability

After a thorough search of the literature and a strict set of inclusion criteria, 13 studies detailing anterior hyoid and laryngeal displacement were analyzed and synthesized for mean displacement measures along with 95% confidence intervals. Results were plotted and examined. These revealed a high degree of variability across studies for all movement trajectories. Potential sources of this variability are discussed below.

Methodological Sources

The inverse relationship between sample size and variability was repeatedly demonstrated with the data from the Bingjie et al. [26] study. It is plausible that a large portion of the variability in the studies reviewed was related to small sample sizes. However, the problem could also lie in the number of swallows per subject. Seven of the studies reviewed included only two swallows per bolus condition, while the others [21–25] limited their analysis to one swallow per bolus condition. Lof and Robbins [11] recommend that at least three repeated trials of swallowing at each bolus volume should be included in VFSS “in an effort to balance the need to capture the individual variability with the negative effects of radiation exposure.” This recommendation was not followed in any of the studies that were reviewed. Future studies should maximize both the number of participants and the number of swallows per condition to potentially reduce variability.

Measurement techniques can also account for variability across studies. Take, for example, the methodology used to extract hyoid bone displacement. While the majority of studies reviewed tracked the position of the hyoid on each frame throughout the swallow, 5 of the 13 studies obtained distance measures by comparing a single frame at rest to another single frame at maximum displacement [3, 5, 16–18]. The choice of the rest frame itself appears to be susceptible to some variability. Some researchers defined the rest frame to be the moment before the hyoid starts moving [17, 18], while others defined it based on the bolus’s location in the mouth/pharynx [5]. Others do not define the rest frame at all [3, 16]. This method of deriving displacement measures from single frames at rest and maximum displacement assumes that the frame of maximum anterior displacement is the same as the frame of maximum superior displacement. This remains to be proven. Furthermore, researchers [21, 29] have shown that the hyoid is

not stable at rest. Importantly, Wintzen et al. [30] showed that the start position and end position of the hyoid were the same for saliva swallows but that the preswallow position of the hyoid lowered with increasing bolus volumes; this was attributed to a lowering of the floor of mouth musculature to accommodate bigger bolus sizes. This finding could single-handedly explain the observed pattern of increased displacement for increased bolus volumes in the studies reviewed.

Another potential methodological source of variability is the choice of the plane of reference used when deriving displacement measures. Based on our inclusion criteria, all of the selected studies rotated the X,Y coordinate system to an anatomical reference. With the exception of one study [21], the Y axis was aligned to the spine via a line drawn through the anterior inferior corners of two cervical vertebrae (most often C2 through C4), with the X axis intersecting perpendicular to Y . Ishida et al. [21] define the X axis via markers on the teeth (occlusal plane), with Y perpendicular to X . Work by Nakane et al. [31] compares different planes of reference in young and elderly subjects. They suggest that Camper's plane (similar to the occlusal plane: an X axis defined by points on the nose and tragus) is the preferred method because it is less susceptible to morphological changes of aging (i.e., osteophytes on the spine) and does not require markers to be fixed to the dentition. Differences in anatomical references may account for some variation in hyoid and laryngeal movement trajectories, especially in elderly subjects.

Statistical Sources

Given that three or more swallows per bolus condition is preferred for capturing representative swallow performance [11], the statistical handling of repeated measures can also impact the manifestation of variability. The representation of variability will change if repeated measures are averaged for a participant or if each swallow is weighted equally in the analysis. Of course, those studies with only one swallow per bolus condition are exempt from this statistical dilemma. Critical review of the seven studies with two swallows per condition reveals that not all analyses were conducted in a consistent manner. Some groups weighted each swallow equally in the analysis [18–20], while others averaged repeated measures at each volume/condition [3, 5, 16, 17]. Kim and McCullough [32] recently proposed that researchers should “investigate biomechanics by analyzing each swallow independently, rather than averaging swallows.” Meanwhile, Max and Onghena [33] provide an important tutorial in the *Journal of Speech Language and Hearing Research* on the proper handling of the “experimental unit.” Contrary to Kim and McCullough's suggestion, they emphasize that repeated measures should be

treated as a single case or unit and failure to do so can result in incorrect statistical conclusions. Once again, variability can manifest from this kind of distinction.

Stimulus Choice

Seven of the studies reviewed [3, 16, 21–25] provide details regarding the weight/volume (w/v) of the contrast medium that they used. A historical study by Dantas et al. [34] showed that the magnitude of maximal anterior hyoid displacement was significantly greater when using high-density barium compared with low-density barium. Interestingly, this fact could explain the higher values for anterior hyoid displacement in Fig. 1 between the Dodds study, which used 250% w/v (high-density), and the Dantas study, which used 40% w/v (low-density).

In a typical VFSS, the clinician cues the patient to swallow with a verbal command. Recent research by Daniels et al. [35] has demonstrated that the use of command swallows significantly reduces swallowing event durations. The effect of cueing on kinematic aspects of swallowing is not yet understood, but it is plausible that the hyoid may be partially pre-elevated during an intentional oral hold of the bolus; this might have the effect of reducing displacement measures, depending on the method used to extract displacement measures. None of the studies reviewed provided details regarding the use of verbal cueing; this element should be controlled in future research.

Patient Sources

Those studies that include gender in their analysis [19–21] provide evidence that variation in superior and anterior hyoid and laryngeal displacement exists for both men and women. It remains unknown whether the magnitude of variation is equal across men and women and requires future in-depth analysis.

Mays et al. [36] examined the relationship between the Frankfort Mandibular Plane Angle (FMA, a measure of craniofacial morphology) and hyoid bone displacement. They found an inverse relationship, i.e., that the greater the FMA, the smaller the anterior hyoid displacement. None of the studies reviewed provided details regarding FMA. The FMA could potentially influence the variability seen in measures of anterior hyoid displacement.

The influence of patient height on kinematic measures of swallowing has been considered by some researchers. For example, Logemann et al. [19, 20] measured the C2-C4 distance as a proxy for height and used this measure as a covariate when comparing groups (i.e., young versus old). However, in the studies reviewed, height was not factored into the mean values and may have had a significant impact on observed variability. It remains unknown if taller

participants would have increased hyoid displacement. Future work should examine whether patient height can account for some of the variability observed in swallowing kinematics.

The criteria by which participants are described may also be a source of variability. Take, for example, the definition of “healthy” subjects in the studies discussed above. Some define healthy subjects as those referred for an assessment but with no evidence of dysphagia [3, 17], whereas others use “healthy volunteers” recruited from the community. It is unclear whether these groups can be expected to behave in the same way and this may be another potential source of the variability.

Lof and Robbins [11] hypothesized that patients with dysphagia may present with significant differences in variability (either reduced or exacerbated), while their average values could appear identical to those of healthy individuals. There is a relative dearth of literature detailing swallowing kinematics in patients with dysphagia. However, in recent years, pioneering work describing hyoid and laryngeal displacement in patient populations has been conducted [22, 25, 26, 29, 32]. Unfortunately, these studies were conducted on relatively small, heterogeneous (in terms of presentation of dysphagia) samples making them difficult to compare across studies. The impact of type and severity of dysphagia on swallowing physiology variability is not yet clearly understood.

Finally, after all the other sources of variability have been considered and controlled for, it is plausible to assume that some portion of the variability can be attributed to variability that is inherent to an individual’s swallow.

Conclusions: Implications for Future Studies

Based on a careful review of the literature and compilation of data on swallowing physiology across several studies, evidence of variability on hyoid and laryngeal displacement emerges in both anterior and superior planes of movement. For researchers, the impact of variability on statistical analysis and normative data development must be seriously considered. Clinicians should recognize the potential of variable patient performance in VFSS on management decisions. Various statistical, methodological, stimulus-related, and patient-related factors are thought to impact the degree to which this variability is apparent. Future work is needed to understand the relative contributions of each of these variables. A large-scale study that carefully controls these issues may provide researchers and clinicians with an accurate picture of variability in swallowing function to inform management decisions, treatment planning, and effect size calculations.

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