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Quantification of vocal tract configuration of older children with Down syndrome: a pilot study

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Objective: To quantify the vocal tract (VT) lumen of older children with Down syndrome using Acoustic Reflection (AR) technology.

Design: Comparative study

Setting: Vocal tract lab with sound-proof booth

Participants: Ten children (4 male and 6 female), aged 9-17 years old diagnosed with Down syndrome. Ten typically developing children (4 male and 6 female) matched for age, gender, and race.

Intervention: Each participant's vocal tract measurements were obtained by using an Eccovision Acoustic Pharyngometer.

Main outcome measures: Six vocal tract dimensional parameters (oral length, oral volume, pharyngeal length, pharyngeal volume, total vocal tract length, and total vocal tract volume) from children with Down syndrome and the typically developing children were measured and compared.

Results: Children with Down syndrome exhibited small oral cavities when compared to control group ($F(1, 18) = 6.55, p = 0.02$). They also demonstrated a smaller vocal tract volumes ($F(1, 18) = 2.58, p = 0.13$), although the results were not statistically significant at the 0.05 level. Pharyngeal length, pharyngeal volume, and vocal tract length were not significantly different between the two groups.

Conclusion: Children with Down syndrome had smaller oral cavities, and smaller vocal tract volumes. No significant differences were found for pharyngeal length, pharyngeal volume, and vocal tract length between these two groups.

Keywords: Vocal Tract Configuration, Down syndrome, Acoustic reflection, Oral cavity

1. Introduction

Down syndrome is the most common genetic disorder related to mental retardation, and occurs in approximately 1 in 800-1000 infants [1,2]. The disorder is caused by a trisomy of chromosome 21, resulting in a particular constellation of physiological and cognitive symptoms, such as intellectual disability, cranio-facial abnormalities, conductive hearing loss, and muscle hypotonia (flaccidity) [2-3]. Research has consistently shown that children with Down syndrome demonstrated particular deficits in speech and language development [4-7]. Expressive language skills were particularly compromised, affecting verbal expression, phonological development, articulation, voice quality, and overall speech intelligibility [8-10].

Children with Down syndrome had a significantly higher prevalence rate of articulatory impairments when compared to typically developing children or those with other developmental disabilities [11]. Barnes *et al.* [12] found that in comparison to children with congenital Fragile X syndrome, children born with Down syndrome made significantly more errors on speech function tasks as a result of oral-motor difficulties. They also found that motor impairments related to Down syndrome had a profound effect on coordination and timing of articulatory movements. The participants in the study demonstrated particular difficulties executing rapid alterations of speech movements required for producing multi-syllabic words [13].

Structural abnormalities of the vocal tract in this population were extensively reported. Westerman *et al.* [14] reported that the palatal dimensions of patients with Down's syndrome were narrower in width, shorter in depth, and lower in height. Shapiro [15] and Uong *et al.* [16] suggested that Down syndrome was associated with

dramatically shorter palatal lengths. Panchon-Ruiz *et al.* [17] demonstrated slightly shorter palatal length in Down syndrome patients. Skrinjaric *et al.* [18] reported that patients with Down syndrome displayed significantly higher frequency of shelf-like or "stair palate" than controls. These structural deviations could have contributed to the speech deficits observed in this population [9,12].

Various research studies have indicated a relationship between structural anomalies and oral-motor impairments for people with Down syndrome. In a study, [19] three pairs of twins, ages 11 and 27 months, were studied and the findings indicated that the children with Down syndrome exhibited a higher incidence rate of oral motor impairments, including excessive tongue protrusion, inadequate lip closure, and uncoordinated jaw function. A study found the general motor deficits in children with Down syndrome impacted the ability to produce the "rapid, alternating movements and timing" required to produce accurate speech [13(p233)].

It has long been expected that people with Down syndrome expressed a different vocal tract configuration. Even today, speech disorders associated Down syndrome were assumed to be attributed to macroglossia (enlarged tongues), which resulted in tongue reduction surgeries (partial glossectomy) to improve speech articulation [20-22]. However, efficacy outcomes for such procedures reflected no improvements in speech as a result of the operations [23-25]. More recent studies have supported an alternate hypothesis, suggesting that macroglossia typically associated with the disorder might be actually the result of a reduced oral cavity creating the appearance of a larger tongue size [16]. This reduced vocal tract volume may contribute to speech difficulties by limiting the distance and range of movements of typical-sized articulators, such as the tongue and velum [20]. In other words, articulatory difficulties are more likely the result of limited tongue movement in the context of a reduced oral

cavity [26]. Smaller anterior facial skeletons and underdeveloped maxillas contribute to a reduced oral cavity volume, and, thus, may affect speech resonance and range of motion of the oral articulators [9,27-29].

Smaller mandibles, missing or poorly developed facial bones, and smaller, wider jaws may indirectly affect speech articulation by creating smaller oral and pharyngeal cavities, thereby impacting speech sounds resonance through the upper airway [30]. Relatively enlarged tonsils and adenoids impinge upon the oral and pharyngeal cavities, further reducing the area of the vocal tract lumen [16]. Similarly, structural abnormalities of the upper airway may also heighten the risk for feeding and swallowing difficulties for very young children with Down syndrome [31-32].

Uong *et al.* [16] used magnetic resonance imaging (MRI) to obtain length, area, and volume measurements of the upper airway and surrounding tissues of children with Down syndrome (age, 3.2 ± 1.4 yr) who had no evidence of obstructive sleep apnea (OSA). By comparing the sequential T1- and T2-weighted spin-echo axial and sagittal images of children with Down syndrome with those of the normal controls, they found that children with Down syndrome had smaller airway volume (1.4 ± 0.4 versus 2.3 ± 0.8 cm³ in controls, $p < 0.005$), causing a smaller mid- and lower face skeleton in this population. They had a shorter mental spine-clivus distance, hard palate length, and mandible volume. Furthermore, the study also demonstrated that the reduced upper airway size in children with Down syndrome was not due to increased adenoid and tonsil size as reported in some previous investigations.

Despite the documented lumen abnormalities associated with Down syndrome, little is known about the oral and pharyngeal dimensional characteristics of these children. The objective of this study was to measure the vocal tract lumen of children with Down syndrome by using Acoustic Reflection (AR) technology (Eccovision

Acoustic Pharyngometer; SensorMedics Corp., Yorba Linda, CA). Specifically, the purposes of this study were to 1) quantify the vocal tract configurations of children with Down syndrome; 2) compare this data against that of typically developing children to determine if significant differences are observed between these two populations; 3) locate specific structural differences along the vocal tract, thereby indicating a potential relationship to speech and/or swallowing difficulties observed in children with Down syndrome; and 4) establish preliminary data of the vocal tract configuration for this population. The study applied non-invasive and cost-effective AR technology that was quite suitable for similar studies that involved large numbers of participants, particularly older children with different types of disorder.

2. Methods

2.1 Participants

This study compared the area-distance curves representing the upper airway of children with Down syndrome against the typically developing controls. The study included 10 older children (4 male and 6 female) diagnosed Down syndrome, aged 9-17 years old. The typically developing controls included 10 children (4 male and 6 female) matched for age, gender, and race. The matching of age, gender and race was important given the large range of ages of older children involved in the investigation, as well as possible gender and race related differences in the VT of older children [33,34]. The participants were recruited from disability advocacy groups in the Portland metro area. Normative data was obtained from a data set used in a previous study [33,35]. using the same procedures and study criteria. All participants were screened for the absence of the following: (1) co-morbid structural anomalies, such as cleft lip or cleft palate; (2) co-morbid neurological impairments; (3) upper respiratory infection at the time of

testing; (4) and previous oro-maxillo-facial surgeries. All participants passed a pure-tone threshold hearing screening bilaterally at 30 dB (HL) for 500Hz, 1,000 Hz, and 2,000 Hz, and 4,000 Hz. Although the normative growth patterns of Down syndrome children are different from those of typically developing children [36], the six VT parameters measured in this study were anatomical in nature, and were highly correlated with body size [37,38], thus attempts were also made to control the homogeneity of body sizes. A one-way ANOVA revealed no statistical significance for weight ($F(1, 18) = 1.32, p = 0.27$), body mass index ($F(1, 18) = 0.012, p = 0.91$) and height ($F(1, 18) = 4.21, p > .05$) between the two groups. Table 1 listed the demographic information for the study groups. (Table 1 to be inserted here.). The Institutional Review Board of Portland State University reviewed the study protocol before data collection commenced, and oral and written informed consent was obtained from the parents or legal guardians of all the participants according to the Declaration of Helsinki.

2.2 Testing Equipment: Acoustic Reflection (AR) Technology

Pharyngometric measurements were obtained using an Eccovision Acoustic Pharyngometer™. This device consisted of two microphones and one sound generator mounted on a 30-cm-long, 1.89-cm inner diameter wave tube, and a microcomputer equipped with digital-to-analog and analog-to-digital converters for software and data processing. The use of AR technology has been demonstrated as a viable option for delineating the parameters of the human upper airway [39-44]. This procedure uses acoustic energies, which are transmitted through a tube into the airway. A fraction of the acoustic wave is reflected back at each point of discontinuity in the upper airway and is recorded by a microphone attached to the mouthpiece [43]. The other end of the

transmitting tube is connected with the CPU that transforms the wave signal with algorithms into dimensional values shown on the monitor (see Figure 1). The device underwent an automatic self-calibration as set by the manufacturer each time a participant was tested. The cross-sectional area of the vocal tract as a function of the distance from the lips to the glottis was plotted (see Figure 2) according to the amplitude and arrival times of acoustic returns. The measured area–distance curves through mouth breathing, that correspond to major vocal tract morphological marks were selected for analysis according to the following criteria: (a) the oral pharyngeal juncture (OPJ that refers to the velum area demarcating oral cavity and pharyngeal cavity) of the mouth-breathing curve best matched the OPJ of the nose-breathing curve, and (b) the curve fluctuated with the smallest magnitude due to airflow changes. The resultant volume–distance relationships were divided into two sections by hand-marking to separate the oral cavity from the pharyngeal cavity as outlined by the manufacturer, using the following standard criteria: an oral region extending from the incisors to the anterior margin of OPJ and a pharyngeal region extending from the oral pharynx to the end of hypo-pharynx (the glottis) (Figure 3). Six measured volume–distance curves were obtained for each participant: oral volume, pharyngeal volume, vocal tract volume (in cubic centimeters), oral length, pharyngeal length and vocal tract length (in centimeters).

2.3 Testing Procedures

In accordance with manufactures guidelines, each participant was tested three times using the following procedure:

1. All subjects were tested while sitting upright in a chair while demonstrating good postural control and head support during normal tidal breathing.

2. A new, sanitized mouthpiece was selected for each participant and sized for optimal fit to prevent air leakage. The wave tube was positioned so that it was positioned parallel to the ground, creating a straight line to the pharyngometer.
3. Subjects were asked to focus on a certain point in space, which was indicated by a picture the researcher attached to the wall. The researcher assisted the participant in sitting up straight and remaining still. The participant was then prompted to think silently of an “oooh” sound to relax the facial muscles, bring the tongue to a neutral position, and close the velum thereby preventing air leakage through the nasal cavity.
4. The clinician elicited three curves during normal mouth breathing. For those children who had difficulty of maintaining mouth breathing, the clinician would use index finger and thumb to approximate their nasal cavities towards the nasal septum to seal nasal breathing. The calculations of these curves were averaged for each participant across the six VT parameters. Note: for one participant, only one valid trial was obtained; consequently, the values for this participant were not based on an average of three trials.

3. Results

The group means and standard deviations of the oral length, oral volume, pharyngeal length, pharyngeal volume, total vocal tract length, and total vocal tract volume for typically developing children and children with Down syndrome were listed in Table 2. (Table 2 to be inserted here). A one-way ANOVA test with Down syndrome as the independent variable and the six vocal tract parameters as the dependent variables was performed to determine if there were significant differences in the vocal tract parameters between the two groups. Significant differences in vocal tract

dimensions of participants in the experimental group and the control group were found in oral volume ($F(1, 18) = 6.55, p = 0.02$), suggesting that children with Down syndrome exhibit small oral cavities when compared to their typically developing cohorts. Additionally, children with Down syndrome demonstrated a trend toward smaller vocal tract volumes ($F(1, 18) = 2.58, p = 0.13$), although these results were not statistically significant at the 0.05 level. Pharyngeal length, pharyngeal volume, and vocal tract length were not significantly different between the two groups. (Figure 4 to be inserted here).

4. Discussion

AR technology has been extensively used as an objective diagnostic tool for locating structural and functional abnormalities within the oral and pharyngeal cavities, used in a variety of clinical applications, including 1) locating site and severity of vocal tract obstructions for patients with sleep apnea [45,46]; 2) establishing degree of nasal airway abnormalities [47,48]; and 3) determining optimal endotracheal tube positioning [41], and 4) documenting changes in the vocal tract due to race and aging [33-35,49]. AR can be a valuable alternative to other objective diagnostic techniques, such as MRI, fluoroscopy, or ultrasound, for assessment of the vocal tract, as the procedure is completely non-invasive and does not expose the participants to radiation. Comparative studies by D'Urzo *et al.* [50] and Marshall *et al.* [51] found that AR measurements could be used in lieu of MRI and CT for quantifying some aspects of vocal tract configurations.

The current results indicated that children with Down syndrome had different vocal tract configurations from those of typically developing children. Particularly, this group may have smaller oral cavities compared to their normally developing cohorts.

This finding has direct clinical implications for speech-language pathologists and other allied health professionals in assessing and treating speech and swallowing difficulties for this population. Previous studies have indicated a correlation between Down syndrome and enlarged tongue size; however, others argued that tongue size maybe normal though it appeared larger within the context of a smaller oral cavity in Down syndrome patients [20]. Uong et al. [16] used MRI to quantify the sizes of major articulators and they found that Down syndrome children had similar tongue sizes with the general population, but smaller mid- and lower face skeleton. Such anatomical reports were in accordance with the findings of the present investigation. Macroglossia (enlarged tongues) among Down syndrome children may not be the “standard” feature of this population. Although larger numbers of Down syndrome participants and typically developing controls with smaller age disparities are clearly needed for future studies in order to anatomically confirm the smaller oral cavities among Down syndrome children, these pilot data logically raised serious questions and concerns on the continuous use of partial glossectomy as a therapeutic modality for Down syndrome patients.

The current results showed no differences in the pharyngeal length and volumes between Down syndrome and normal groups. Since most of the intrinsic lingual musculatures do not extend into the pharyngeal cavities, reduction of lingual motility and restricted range of motion of the oral articulators may contribute, to some extent, to the articulatory difficulties associated with the disorder. A smaller oral cavity together with typical-sized oral articulators may inhibit the coordination of the rapid, alternating movements of the articulators required for speech, thereby reducing intelligibility [13]. Such articulatory differences may also contribute to impairments in speech-motor development. Structural differences “are likely to influence

speech-motor development and consequently negatively impact the articulatory and phonatory abilities of children with Down syndrome” [10(p301)]. Since function and structure are intrinsically reciprocal to each other, the restricted movements of the articulators can also contribute to the development of structural differences in the Down syndrome population. This is illustrated by the findings that there were more similarities than differences between Down syndrome children and their typically developing younger cohorts among younger children than older children [27,28].

Differences in vocal tract volumes are also correlated with changes in vocal qualities. A recent line of research found that volumetric sizes of the vocal tract could be linked to acoustic differences across different racial groups, as well as different age groups [33-35]. These findings indicated some limitations of the traditional tube-resonator model in which vocal tract length has been used as the primary factor in predicting changes in formant frequencies. As the authors stated [35(p638)], “morphological differences (especially vocal tract volumetric parameters) are partially responsible for formant frequency differences”. Other studies have further supported the hypothesis that volumetric differences of the vocal tract affect vocal quality. For instance, Leddy [26(p71)] concluded that structural differences observed in Down syndrome, “may create a smaller mouth and throat, which may influence how speech sounds travel through those spaces and how vocal quality is produced”.

Differences in oral cavity volume may also be related to feeding and swallowing difficulties in infants with Down syndrome. A smaller oral cavity can result in difficulties with sucking, swallowing, and salivation [32]. Decreased sucking pressure during feeding associated with Down syndrome may be exacerbated by reduced oral cavity volume by causing the tongue to impinge upon the back of the mouth, thereby inhibiting the coordinated, peristaltic tongue movements required for

proper feeding [52]. Furthermore, decreased oral cavity could contribute to an open-mouth breathing posture during feeding, which is disadvantageous for maintaining sufficient sucking pressure during feeding [52,53]. Restrictions in lingual motility as a result of a smaller oral cavity may induce oral-phase swallowing difficulties with regard to mastication, bolus formation, and oral transit times [52]. Understanding of these factors is critical for speech-language pathologists in devising appropriate feeding interventions especially for Down syndrome children at very young ages.

This pilot study was clearly limited by the small number of participants and the large age disparities among the participants. Due to the difficulties of some participating Down syndrome children to follow the examiners' directions of mouth breathing during pharyngometric recording, the testing procedures were not exactly consistent across all participants. However, the investigation demonstrated that AR technology could be used for large scale comparative studies of vocal tract configurations when it is not feasible to use MRI and other conventional imaging technologies. The findings of the study motivated the speech pathologists to develop new therapies with the aim of enhancing the compromised mobility of the articulators within smaller oral cavities of Down syndrome children. It also raised legitimate concerns for the continuous use of partial glossectomy for treating children and adults with Down syndrome.

Clinical Message

Children with Down syndrome may have smaller oral cavities, and smaller vocal tract volumes when compared to the typically developing children. Therefore, partial glossectomy as a rehabilitation treatment for this population is not clinically justified.

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Captions:

Figure 1. Schematic Chart of Pharyngometer.

Figure 2. 3-D Illustration of Vocal Tract.

Figure 3. Area-distance curve of vocal tract dimensions from a pharyngometer. Note: Pharyngeal Cavity is calculated by combining Oral Pharynx and Hypo-pharynx.

Figure 4. Average oral length (OL), pharyngeal length (PL), vocal tract length (VTL), oral volume (OV), pharyngeal volume (PV), and vocal tract volume (VTV) for children with Down syndrome (DS) and typically-developing children (TD).

Table 1. Demographic data for children with Down syndrome and the typically developing controls.

	Down syndrome		Typically Developing	
	Male (n = 4)	Female (n = 6)	Male (n = 4)	Female (n = 6)
Age (yrs)	11.25	13.83	11.25	11.25
SD	2.87	3.19	2.87	3.19
Range	9 -15	9 - 17	9 – 15	9 - 17
Height (cm)	141.75	138.17	148.00	155.33
SD	19.19	11.50	15.03	10.10
Range	122 - 168	127 - 153	135 – 160	135 - 173
Weight (kg)	39.75	45.67	51.00	52.33
SD	12.20	15.55	25.53	14.35
Range	29 - 57	34 - 76	34 – 88	34 - 72
Body Mass Index (kg/m²)	19.43	23.23	22.53	21.6
SD	.929	6.37	7.92	4.83
Range	18.1 - 20.2	18 - 32.5	18.3 - 34.4	17.6 - 28.7

Table 2. Means (*m*) and Standard Deviations (*SD*) of oral length (OL), oral volume (OV), pharyngeal length (PL), pharyngeal volume (PV), vocal tract length (VTL) and vocal tract volume (VTV).

	Down syndrome				Typically Developing			
	Male		Female		Male		Female	
	<i>m</i>	<i>SD</i>	<i>m</i>	<i>SD</i>	<i>m</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
OL (cm)	7.43	0.39	8.08	0.13	7.75	0.56	7.62	0.18
OV (c.c)	31.79	7.42	34.78	3.56	38.71	3.84	40.49	3.58
PL (cm)	9.00	0.49	9.11	0.62	9.23	0.11	9.32	0.22
PV (c.c)	15.28	5.56	19.81	3.41	15.82	4.84	23.83	10.13
VTL (cm)	16.52	0.48	17.19	0.57	16.71	0.44	16.75	0.28
VTV (c.c)	46.83	11.71	54.62	6.61	54.21	2.87	62.91	13.85

Figure1
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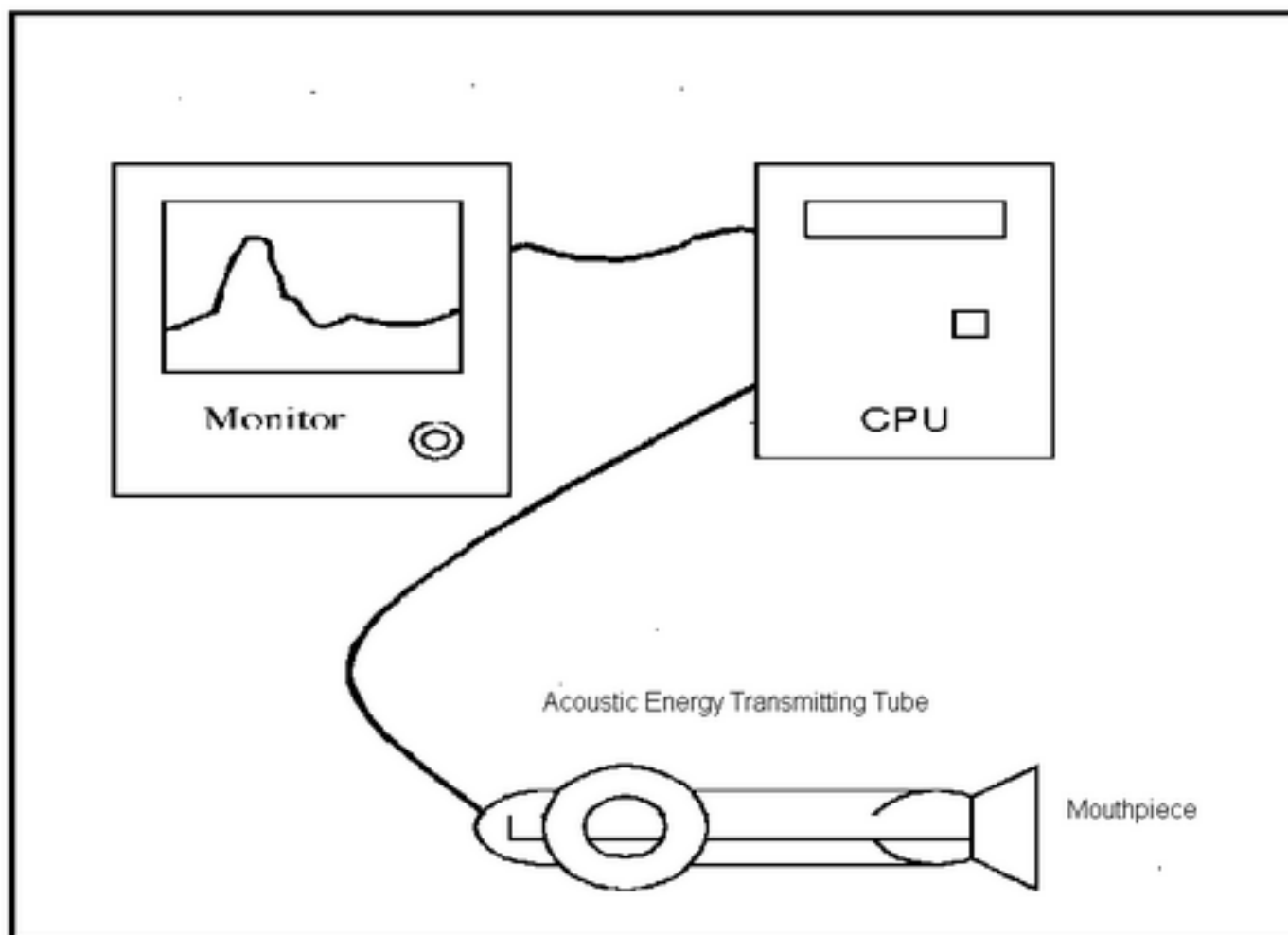


Figure2

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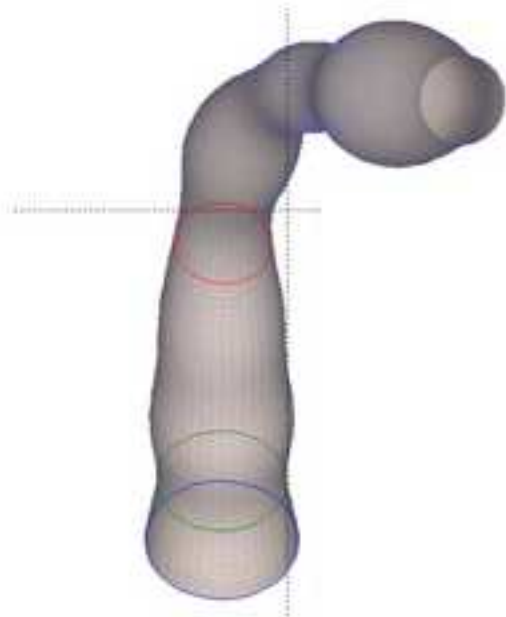


Figure3

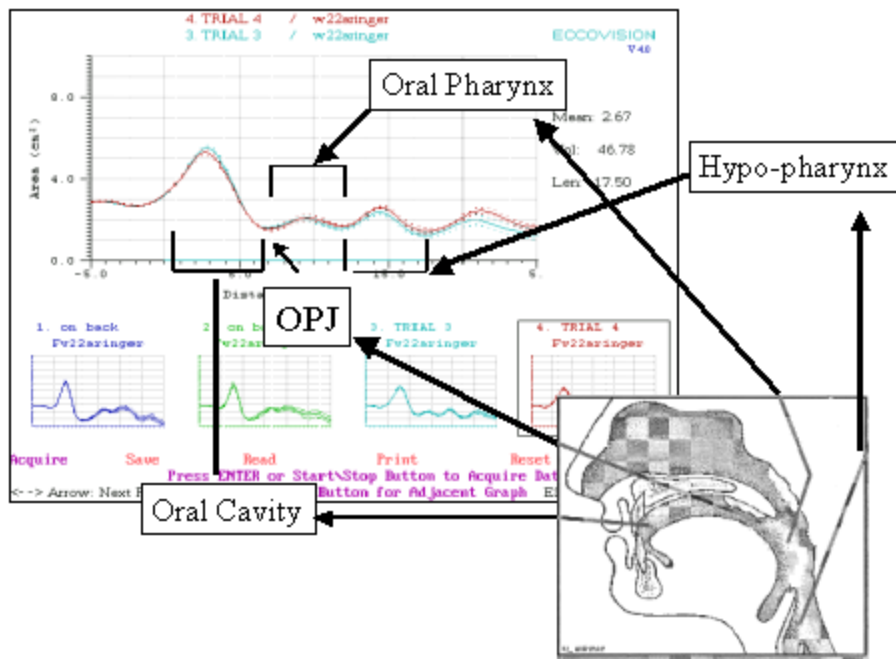


Figure4

