

DYSPHONIA SEVERITY INDEX IN CHILDREN WITH VELOPHARYNGEAL DYSFUNCTION: A PRE-POST OPERATIVE COMPARISON.

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ABSTRACT

Background & Objective: Voice is an important tool for communication through which we express our thoughts in form or sounds produced by movement of vocal folds. The voice quality is affected in children with cleft lip and palate due to velopharyngeal dysfunction (VPD) which is associated with abnormalities of velum leading to hyper adduction of vocal folds. Dysphonia Severity Index (DSI) is an objective multiparametric approach to evaluate voice quality. The present study aimed to compare the Dysphonia severity index (DSI) in children with velopharyngeal dysfunction before and after velopharyngeal surgery.

Method: Twelve children (6 males and 6 females) with Velopharyngeal dysfunction in the age range of 7-12 yrs were considered for the present study. Individuals diagnosed to have velopharyngeal dysfunction by craniofacial team using cineradiography were considered for the study. Maximum phonation time (MPT), frequency and intensity, jitter measurements were made using Lingwaves voice clinic suite pro software Version 2.5 (Wevosys, Germany).

Results and conclusion: There was significant difference between children with velopharyngeal dysfunction and age matched typically developing children on Dysphonia Severity Index values. There was a significant difference for I-low ($p=0.03$) and Dysphonia Severity Index ($p=0.01$) for pre and post-operative conditions. There was significant difference between gender on F_0 high and females (3.11) had better DSI values compared to males (2.37). These results of the present study suggest the need for gender specific voice therapy goals in rehabilitation of voice problems in children with VPD.

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INTRODUCTION

Speech is a vocalized form of human communication that is dependent on the organic integrity of the central nervous system, structures and function of the organs that comprising the speech production mechanism of the human body. The speech production process can be divided into three parts namely respiration, phonation and articulation. Respiration provides the exhalatory air supply needed to produce the speech sounds. Phonation is concerned with the vibratory mechanism that is needed to change the air supply into voiced speech sounds. Articulation is concerned with shaping sounds into specific phonemes of a language.

Velopharyngeal dysfunction (VPD) is an inability to completely close the velopharyngeal part of the oral cavity during speech. This result in the escape of air into the nasal cavity during speech causing hypernasal vocal resonance and nasal emission. The causes of VPD may result from congenital short palate, deep pharynx and malinsertion of the levator muscles [15]. Individuals with improper functioning of their velopharyngeal mechanism can show

disorders of resonance, articulation and voice. Voice problems refer to disordered phonation at the level of the larynx and can include hoarseness, breathiness, low volume, and/or abnormal pitch [15]. Though the larynx is the primary structure for voice production, this system also requires the integration of the respiratory system and the oral and nasal cavities of the vocal tract. Due to the integrated nature of the speech system, problems at the level of the velum may affect the functioning of the larynx.

High prevalence of voice problems in children with velopharyngeal dysfunction (VPD) were reported in literature [2, 3, 14, 19]. The authors hypothesized that individuals with velopharyngeal disorders use grater or hyper adduction of vocal folds to compensate the inadequately functioning velopharyngeal closure. Voice quality refers to those voice characteristics that recognize an individual and to differentiate that individual from the others. The abnormal voice quality in children with VPD may result from forced use of vocal folds in order to

compensate the inappropriate loudness produced by inadequate closure of velopharyngeal closure.

The measurement of voice quality is important for grouping or describing the problem and plan for management. The perceptual and instrumental methods were used to evaluate voice quality in individuals with velopharyngeal dysfunction. The perceptual evaluation methods were widely used for documentation of severity in voice quality and the major drawback is that it is subjective and has a very low reliability. The objective measures of voice quality assess various acoustic parameters of voice. The previous studies reported that not all the acoustic parameters of voice correlate well with the perceived voice quality [7,8,16]. A multiparametric approach was developed which uses a combination of several acoustic and aerodynamic parameters to better correlate with the perceived voice quality.

Dysphonia severity index (DSI), one of the multiparametric approaches for objective measurement of voice quality was developed by Wuyts et al. [23]. The author considered several acoustic and aerodynamic parameters such as Jitter (%), Shimmer(%), Noise to Harmonic Ratio (NHR), Highest frequency (F₀-High) (Hz), F₀-Low (Hz), F₀-Range (Hz), Semitone-range, Lowest Intensity (I-Low) (dB), I-High (dB), I-Range (dB), maximum phonation time (MPT, s), Vital Capacity (VC) (cc) and Phonation Quotient (PQ) (cc/s) to calculate the weightage of each parameter on perceived voice quality. On analyzing these entire variables on normal and disordered population, the author derived the index consisted of weighed parameters such as highest fundamental frequency (F₀ -high), lowest intensity (I-low), maximum phonation time (MPT) and jitter (%). The DSI is constructed as $DSI = 0.133 * MPT + (0.00533 * F_0\text{-High}) - (0.263 * I\text{-Low}) - (1.183 * Jitter \%) + 12.4$.

The resulting DSI values vary between $\geq +5$ (No dysphonia) and < -5 (severe dysphonia). Since the range of possible scores on the separate parameters is wide, scores + 5 (good voice quality) or - 5 (poor voice quality) are possible as well (Wuyts et al., 2000). DSI is not limited to the interval +5, -5. In clinical practice values of -6 and more are also reported. This is generally caused by high jitter values. The DSI can be obtained easily and quickly by speech pathologist in a clinical setup. The DSI is very useful in evaluation of individuals with voice problems.

Van Lierde et al. [19] examined the vocal quality and effect of vocal quality on gender in children with cleft lip and palate. Twenty eight children with unilateral or bilateral cleft lip and palate were considered for the study. The voice quality was measured using the using videolaryngostroboscopic and perceptual evaluations, aerodynamic, voice range, acoustic, and dysphonia severity index (DSI) measurements. The results showed gender related vocal quality differences, the male children showed over all vocal quality of +0.62 with slighter degree of hoarseness and female children showed +2.4 reflecting a perceptually normal voice. The results of the present study provided valuable insights into the vocal quality characteristics children with cleft palate.

Van Lierde et al.[22] studied speech outcome on voice characteristics in seven subjects in the age range from 4.7 to 9.1 years with a mean age of 6.9 years postoperatively following pharyngeal flap surgery. Dysphonia severity Index (DSI) was calculated in subjects postoperatively after one year. The stroboscopic evaluation

for vocal outcome showed normal vocal folds. The results showed that overall vocal quality of the DSI was 1.7 (range 0–4.8) reflecting, as very slightly impaired vocal quality. These results may be hypothesized due to the stronger adductory force on the vocal folds to minimize hypernasality and to reach specified voice intensity.

These studies by Van Lierde et al.[19, 22] in literature highlights the incidence of voice problems in children with cleft lip and palate using DSI. The authors used various methodologies but there are no studies in literature comparing the pre-operative DSI parameters with post-operative DSI in individuals with velopharyngeal dysfunction. Hence the present study is aimed to compare the pre-operative DSI scores with post-operative scores after surgery for velopharyngeal closure. The present study is also hypothesized to study the effect of gender on vocal quality in individuals with velopharyngeal dysfunction.

METHOD

Participants

Twelve children (6 males and 6 females) with cleft lip and palate in the age range of 7 to 12 yrs who have undergone primary palatal repair (cleft of hard palate and soft palate, cleft of the soft palate) for the closure of the cleft will be identified after consultation with plastic surgeon were considered for the present study. All the participants were evaluated by the craniofacial team at unit for structural oro-facial anomalies (U-SOFA), AIISH. Individuals diagnosed to have velopharyngeal dysfunction by craniofacial team using direct visualization procedures such as cineradiography/ nasoendoscopy with the help of plastic surgeon and radiologist were considered for the study. All the subjects underwent secondary speech surgery under the same surgeon and the details were represented in table 1. None of them had cleft associated with syndromes, cognitive deficits, neuromotor dysfunction, and a hearing threshold above 20 dB in both ears.

Table 1. Demographic details of children with VPD

S.No	Subject	Age/Gender	Type of Surgery
1	A	9yrs/M	Furlow's Z plasty
2	B	9yrs/M	Furlows Palatoplasty
3	C	7yrs.M	Furlows Double opposing Z plasty
4	D	12yrs/M	Furlow's Z plasty
5	E	7yrs/M	Furlows Palatoplasty
6	F	7yrs/M	Furlows Palatoplasty
7	G	9yrs/F	Furlow's Z plasty
8	H	12yrs/F	Hynes Pharyngoplasty
9	I	7yrs/F	Hynes Pharyngoplasty
10	J	8yrs/F	Furlow's Z plasty
11	K	11yrs/F	Furlow's Z plasty
12	L	7yrs/F	Furlow's Z plasty
Mean age : 8.75 years			

All the subjects were followed up after surgery and evaluations were done after six months. A written consent was obtained from the parents of the participants and they were explained about the method and the procedure of the study.

Procedure and Instrumentation

The individuals considered for present study were evaluated for voice quality using Lingwaves voice clinic suite pro software Version 2.5 (Wevosys, Germany).The Lingwaves software is a computer based standardized measurement system for voice and speech diagnostics. The parameters used for DSI measurements are the highest fundamental frequency (F₀-high in Hz), lowest intensity (I-low in dB sound pressure level (SPL), maximum phonation time (MPT in sec), and jitter (%). The testing was done in a

very quiet environment with a help of SPL –meter and the sound pressure level during the silence did not exceed more than 45 dB (A). The distance between SPL meter and individuals mouth was about 30 cm and the subject was instructed to keep the position same throughout the procedure. The same procedure was carried out six months postoperatively for each subject. The different parameters of DSI includes

a) Maximum Phonation Time (MPT/sec)

Maximum Phonation Time (MPT) was measured on the basis of three trials with the vowel /a/, sustained at the individual's habitual pitch and loudness in free field. The length of the sustained vowel was measured using Adobe audition software (version 3). The best and longest sustained vowel /a/ of the three trails was measured is seconds (s) and considered for analysis.

b) Highest Frequency (F₀-High/Hz) and lowest Intensity (I – low/dB)

The highest frequency and lowest intensity of DSI was measured using Voice diagnostic centre (VDC) of lingwaves software. The voice diagnostic centre represents a combined voice range profile analysis and voice quality analysis. The subjects were instructed to maintain a distance of 30cm from the sound level meter. They were asked to phonate vowel /a/ as softly as possible at their habitual pitch and later they were asked to phonate the vowel /a/ going up to the highest pitch and coming down to the lowest pitch. The subjects performed three trials and the better one was considered for analysis. The highest Fo and lowest intensity were calculated from phonetogram VDC display.

c) Jitter (%)

Jitter (%) is a period to period variation in fundamental frequency and it was calculated using Vospector –DSI in Lingwaves software. With the lingwaves Vospector we can take the measurements of voice parameters were extracted for calculation of DSI. The subjects were asked to phonate a vowel /a/ at comfortable

pitch and sustain it for 2 to 3 seconds. The middle portion of the recorded phonation more than one second was selected for calculation of jitter (%).

The DSI is constructed as $DSI = 0.133 * MPT + (0.00533 * F_0\text{-High}) - (0.263 * I\text{-Low}) - (1.183 * Jitter \%) + 12.4$. The Lingwave DSI classification is a different compared to Wuyts et al.²³ because the authors used an old jitter algorithm form Kay Elemetrics system. The lingwaves uses a newer evaluated clinical jitter algorithm with on average higher values (<-2.0 –Severe aphonia, -1.9 to +0.3 – Constant dysphonia, +0.4 to +2.2 – Moderate dysphonia ,+2.3 to +3.3 – Slight to moderate dysphonia ,+3.4 to +4.3 – Slight dysphonia , > 4.4 –No dysphonia).

STATISTICAL ANALYSIS

A commercially available IBM SPSS 20 was used for statistical analysis of obtained voice data. Wilcoxon signed rank test was done to find significant difference between the pre-operative and post-operative values of Dysphonia severity index (DSI). Multivariate analysis of variance (MANOVA) was done to find out the effect of gender on the parameters of DSI. To find the test retest reliability 10% of the data were reanalysed using the same software and the results showed greater than 90% reliability.

RESULTS

The results of the present study are explained in the following subsections

a) Comparison of DSI parameters in individuals with VPD across gender for pre and post-operative conditions.

The mean and standard deviation of DSI parameters across gender for both conditions were compared. The data was further compared with normative data for parameters of DSI in typically developing children [9] and the values were represented in Table 2.

Table 2. Mean and S.D for DSI parameters in Individuals with VPD for pre and post operative conditions across gender.

Parameter	Preoperative		Postoperative		Normative Heylen et al. (1998)	
	Males	Females	Males	Females	Males	Females
MPT(Sec)	10.16 (0.75)	10.33 (2.73)	10.16(1.83)	10.33(2.87)	14.23(0.27)	13.36(0.22)
F0-High(Hz)	632.81 (23.53)	740.35(22.10)	636.56 (12.81)	740.80(12.22)	861(35)	901(31)
I-low(dB)	53.72(3.76)	56.16 (2.63)	52.80 (2.57)	53.58(2.87)	48.3(0.5)	47.8 (0.4)
Jitter (%)	1.48 (0.77)	0.47 (0.48)	0.83(0.67)	0.51(0.25)	0.55(0.01)	0.61(0.01)
DSI	1.28(1.50)	2.51 (0.61)	2.37 (1.36)	3.11(0.84)	5.7	6

The results of preoperative conditions showed that males had higher values in maximum phonation time and jitter than female subjects. But females had grater values for Fo-high, lowest intensity, DSI than male subjects. In post operative conditions (after 6 months) the male participants showed grater values for MPT, lowest intensity, Jitter than female subjects. And female subjects showed grater values for Fo-high and DSI values.

When comparing the means scores of Dysphonia severity index (DSI) for males and females across pre and post operative conditions both the genders had increased DSI values compare to their preoperative scores. The preoperative quality of voice was found to be more affected in males (DSI =1.28) representing a moderate degree of dysphonia but in females (DSI =2.51) it was found to be better than males but still had a slight to moderate level of

dysphonia. The post postoperative DSI was found to be better in females (3.11) representing slight or minimal degree of dysphonia and males (DSI = 2.37) had slight to moderate dysphonia. Both females and males had improved voice quality compared to preoperative measurements.

The obtained mean scores for pre and post-operative DSI and its parameters were compared to the normative data by Heylen et al. [9] using Mann Whitney U test. The results showed that there was a significant difference between normals and children with VPD on MPT, Fo-high, I-low and DSI for both pre and post-operative conditions. There was no significant difference was not seen across gender between pre and post-operative conditions.

b) Effect of gender on DSI parameters across DSI values for preoperative condition.

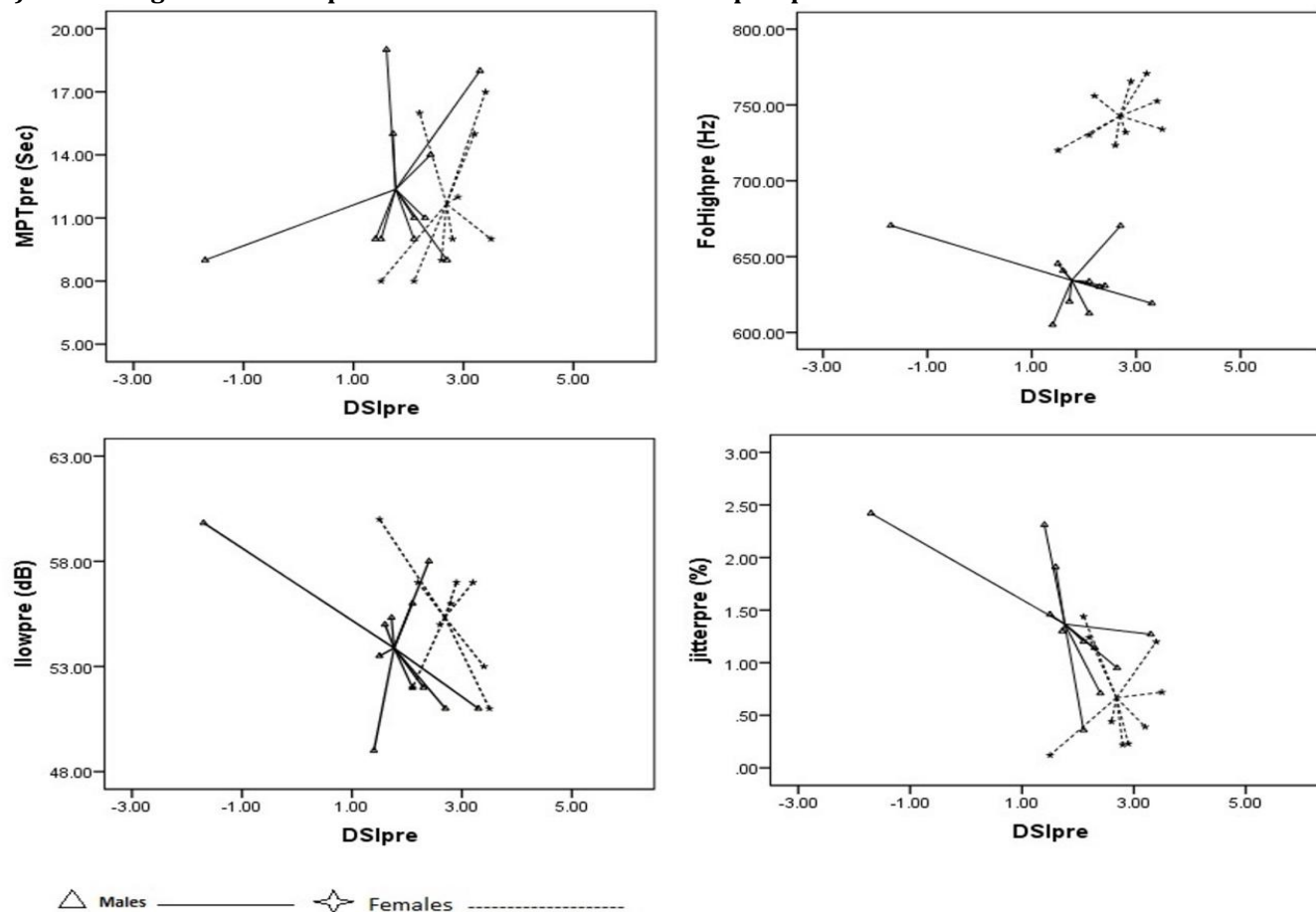


Fig.1. Scatterplot showing effect of gender on DSI parameters A. MPT B.Fo-high C.I-low D. Jitter for preoperative condition.

The figure 1 shows the relationship between the parameters such as MPT, Fo-high, I-low and jitter for males and females across preoperative DSI values. MANOVA was done to find out if there is any significant difference between the gender and DSI parameters. The results showed a significant difference between the gender on F₀-high values [F (1, 16) = 66.54, p < 0.001] and Jitter (%) [F (1, 16) = 7.24, p < 0.05] across gender in preoperative condition. Also there was no significant difference was seen between the gender for parameters such as MPT, I-low and DSI.

c) Effect of gender on DSI parameters across DSI values for postoperative condition.

The figure 2 shows the relationship between the parameters such as MPT, Fo-high, I-low and jitter for males and females across post-operative DSI values. MANOVA was done to find out if there is any significant difference between the gender and DSI parameters. The results showed that there was significant difference between the gender on F₀-High values [F (1, 16) = 207.76, p < 0.001] between both genders in preoperative condition. And there was no significant difference between the gender for parameters such as MPT, I-low, Jitter and DSI.

d) Comparison of Pre and post-operative DSI parameters.

Wilcoxon signed rank test was done to find if there is any significant difference between the preoperative evaluation of DSI and its parameters with post-operative evaluation of the same parameters. The results revealed that for male subjects there was no statistically significant difference for MPT, F₀ -high, I-low, Jitter and DSI across

both pre and post-operative conditions in male subjects. For female subjects significant difference was found for DSI (p < 0.05). For parameters such as MPT, I-low, Fo-high and Jitter there was no significant difference in female subjects across both conditions.

Wilcoxon signed rank test was done by combining male and female subjects to find the overall difference between pre and post-operative conditions in individuals with velopharyngeal dysfunction. The results revealed that there was a significant difference on I low (p = 0.034 < 0.05) and DSI (p = 0.015 < 0.05) between both conditions.

DISCUSSION

The present study is aimed to compare the dysphonia severity index in individuals with velopharyngeal dysfunction before and after surgery and to study the effect of gender on DSI values in both the conditions. The present study investigated twelve children (Mean age =8.75 yrs) with VPD before and after surgery. The DSI, an objective voice quality measures were taken in all the subjects preoperatively and six months after velopharyngeal surgery. The statistical analysis revealed that post-operative DSI scores were better than pre-operative condition in both male and female subjects. But the vocal qualities of females were found to be better than male subjects in both pre and post-operative conditions.

The results of the present study are in consonance with the previous studies that there was an increase in the symptom of hoarseness in individuals with cleft palate compared to normal population. The rate of dysphonia was in individuals with cleft palate is 12 to 43 %

higher than the normal [5,10,13]. The increase in hoarseness in individuals with cleft palate was the laryngeal system trying to compensate for abnormal velopharyngeal valving [12,19].

Individuals with velopharyngeal dysfunction also associated with poor articulation and increased incidence of hypernasality. The poor articulation is often compensated by the use of pharyngeal and glottal sounds. The glottal stops in particular have been indicated in literature to cause hoarseness [3,19]. Hamming, Finkelstein and Sidman [6] explained the cause for voice problem in Individuals with VPD as greater adductory force on the laryngeal structures in order to reduce nasality and reach a certain vocal intensity. This gives the impression that there is no single definite cause for hoarseness in individuals with VPD, suggesting that it is mostly multifactorial in nature.

The individuals with velopharyngeal dysfunction showed an increased jitter, lesser maximum pitch and low intensity compared to normal individuals for both males and females. These results were supported by previous studies [12,20,24] where the individuals with cleft

palate had demonstrated with increased frequency perturbations. The reduced maximum pitch in both the gender also supports the previous studies in the literature [4,18]. Our study supports the previous study by Boone and McFarlane ¹ the suggested that a reduction in Fo can decrease hypernasality.

According to Peterson -Falzone [17] the increased respiratory effort which makes the vocal fold to hyper adduct does not change the intraoral breath pressure in individuals with VPD and it lost through inefficient velopharyngeal mechanism. The decreased voice quality in male participants compared to female subjects in this present study was supported by Van Lierde et al. [20]. These findings enlighten the need for specific voice therapy goals in male participants with velopharyngeal dysfunction. The mean dysphonia severity index for pre-operative condition was found to be 1.90, a moderate dysphonia and six months postoperatively it was 2.74, a slight dysphonia. This was in concordance with the study by Van Lierde et al [22]; he reported an increase in DSI scores postoperatively after VPD surgery.

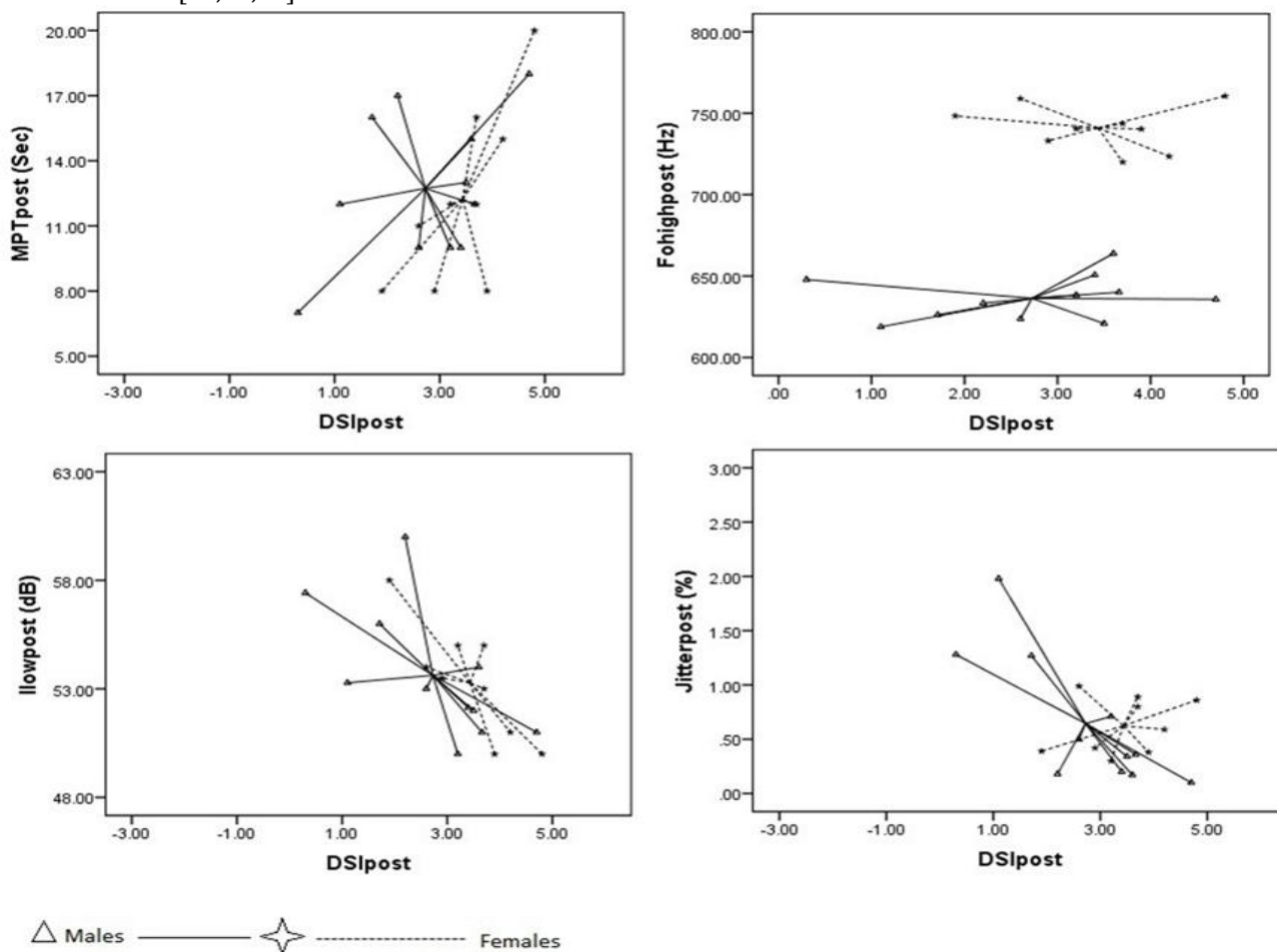


Fig.2. Scatterplot showing effect of gender on DSI parameters E. MPT F. Fo-high G.I-low H. Jitter for post operative condition.

CONCLUSION

The present study investigated DSI and its parameters before and after VPD surgery in individuals with velopharyngeal dysfunction. The study also hypothesised the effect of gender on DSI and its parameters. The results showed that there was a significant difference on DSI between pre and post-operative condition. Although the post-operative evaluation was carried out six months after VP surgery, better voice quality was observed across gender. Future longitudinal

research study is essential for evaluation of voice quality in individuals with VPD to explore the effect of surgery on voice quality in individuals with VPD.

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