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dystonia)

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Utility of maximum phonation time in the

diagnosis of spasmodic dysphonia (laryngeal

Main Article

Nupur Kapoor Nerurkar takes responsibility for the integrity of the content of the paper

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Abstract

Objectives. Maximum phonation time is a simple test used to assess glottic competency. Our objective was to evaluate any correlation between maximum phonation time and spasmodic dysphonia as adductor spasmodic dysphonia and abductor spasmodic dysphonia have an adductor and abductor overdrive, respectively.

Methods. A 3-year data-review was performed for patients diagnosed with adductor spasmodic dysphonia, abductor spasmodic dysphonia and mixed spasmodic dysphonia. Maximum phonation time was noted on the first visit and compared with a control group. **Results.** Average maximum phonation time in adductor spasmodic dysphonia, abductor spasmodic dysphonia and control group was 25 seconds, 9 seconds and 16 seconds. A significant difference was found for adductor spasmodic dysphonia and abductor spasmodic dysphonia. A receiver operating characteristic curve analysis between adductor spasmodic dysphonia and control groups showed a positive predictive value of 81.3 per cent, negative predictive value of 83.9 per cent, sensitivity of 79.6 per cent and specificity of 85.2 per cent. Level of evidence = 4. **Conclusion.** We recommend that maximum phonation time be added to the diagnostic armamentarium of spasmodic dysphonia. This correlation between maximum phonation time and spasmodic dysphonia has not been previously published.

Introduction

Sir Morell Mackenzie defined spasmodic dysphonia in 1868 as "spasmodic action of the tensors, causing the vocal cords to be unduly and irregularly stretched, and consequently giving rise to a voice which is feeble, jerky, unsteady, and constantly rising to a high key." From being described as "hysterical affection" by Ludwig Traube in 1871, the pathophysiology of spasmodic dysphonia today is best understood and elaborated by Simonyan *et al.*, who described it as a "primary focal laryngeal dystonia characterised by loss of voluntary control of vocal fold movements during speech production due to involuntary spasms in the laryngeal muscles". Spasmodic dysphonia is further classified into adductor spasmodic dysphonia, abductor spasmodic dysphonia and mixed spasmodic dysphonia, out of which 29 per cent are associated with vocal tremors, as studied by Tanner *et al.* Laryngeal dystonia is the more currently acceptable terminology for spasmodic dysphonia as it appropriately indicates the true nature of the condition.

Adductor spasmodic dysphonia has a higher prevalence (82 per cent)^{6,7} and is a result of spasmodic bursts of the closing muscles (adductor) of the vocal folds during vowel production, which result in voice breaks.⁸ Vocal tasks, such as speaking sentences which consist of mainly vowels such as "we eat apples all day," highlights this disorder.⁹ Abductor spasmodic dysphonia, a less prevalent form of the disorder, accounts for 17 per cent of spasmodic dysphonia cases and is defined as hyperabduction (uncontrolled opening) of the vocal folds, prolonging voiceless consonants before vowels¹⁰ and can be elicited by disrupted sentences such as, "he is hiding behind the house".⁹ Mixed spasmodic dysphonia is a combination of both adductor spasmodic dysphonia and abductor spasmodic dysphonia¹¹ with a variable component of each type.

Haslinger *et al.* and Ali *et al.* performed neuroimaging studies during voice and narrative speech production in adductor spasmodic dysphonia patients and found activation changes in the laryngeal/orofacial sensorimotor cortex and basal ganglia, ^{12,13} which was further studied by Simonyan *et al.*³ Due to reduced g-aminobutyric acid (GABA) metabolism and dopaminergic receptor binding, excessive motor cortical excitation is seen, resulting in laryngeal dystonia. With better understood pathophysiology of spasmodic dysphonia, recent research has revealed newer modalities of treatment such as use of GABA (Xyrem®) in alcohol-responsive patients. ¹⁴ However, this is currently not commercially available for use in patients and the gold standard for spasmodic dysphonia is still botulinum neurotoxin injection, often given with laryngeal electromyography monitoring. ¹⁵

In order to initiate appropriate treatment, an accurate diagnosis is of paramount importance. Diagnostic challenges in spasmodic dysphonia persist because there is no

© The Author(s), 2024. Published by Cambridge University Press on behalf of J.L.O. (1984) LIMITED objective test available to categorically confirm the same, thus physicians rely on a subjective impression of the patient's voice, particularly in response to various vocal tasks, subjective videostroboscopy and exclusion of other pathologies along with a neurology opinion. A pre-phonatory burst in laryngeal electromyography may be used for the diagnosis of spasmodic dysphonia, however laryngeal electromyography is not routinely performed for diagnosis but used for therapeutic injection of botulinum neurotoxin. The availability of an objective parameter in the diagnosis of spasmodic dysphonia would be extremely useful due to the above-mentioned reasons.

Our aim is to evaluate the utility of maximum phonation time in the diagnosis of spasmodic dysphonia, which is an easily performed objective test. The reasoning behind studying maximum phonation time as a test in the diagnosis of spasmodic dysphonia is the adductory or abductary overdrive present in adductor spasmodic dysphonia and abductor spasmodic dysphonia, respectively, which may consequently alter the maximum phonation time, due to the altered glottic closure pattern. One of the most common differential diagnoses of spasmodic dysphonia is muscle tension dysphonia, which presents with pain on phonation and tenderness over the thyrohyoid membrane. 17,18 In laryngeal isometric muscle tension dysphonia there is an overdrive of the posterior cricoarytenoid muscle (abductor), which results in a posterior phonatory gap. 17 This can explain the reduced maximum phonation time found in muscle tension dysphonia patients. In contrast to this, in adductor spasmodic dysphonia there is an overdrive of the adductor group of muscles, which may lead to an increase in maximum phonation time. Thus, our hypothesis was that due to an adductor overdrive in adductor spasmodic dysphonia the maximum phonation time should be increased and in abductor spasmodic dysphonia, due to abductor muscles overdrive, maximum phonation time should be decreased. Hence, the objective of our study is to evaluate any significance between maximum phonation time in both adductor spasmodic dysphonia and abductor spasmodic dysphonia as compared to a normal control group.

Maximum phonation time is the maximum time in seconds that a person can sustain a vowel produced in a single breath with comfortable pitch and loudness. 19 In spasmodic dysphonia this phonation has multiple spasms and it is essential to count the maximum phonation time as the total duration of this spasmodic phonation, though strictly of one breath. The patient should not be permitted to take a new breath while measuring the maximum phonation time. Thus it is essential to demonstrate and explain clearly to the patient what the test is in order to get accurate and representative recordings. The recording of maximum phonation time only requires a stopwatch, where three such readings are taken, the highest among them is taken as the maximum phonation time for that patient. Maximum phonation time is a quick, easy and zero-resources tool that can be used to assist in ascertaining the glottic closure function of the vocal folds.

Materials and Methods

This study received ethical clearance by the Institutional ethics committee. Our study is both a retrospective and prospective study. Using a database of a voice clinic in a tertiary health care centre, patients diagnosed with adductor spasmodic dysphonia, abductor spasmodic dysphonia and mixed spasmodic dysphonia from January 2020 to December 2022 were reviewed. The diagnosis of spasmodic dysphonia had been

made by subjective voice assessment by hearing the voice, performing vocal tasks, performing flexible videostroboscopy along with vocal tasks, and was confirmed by the response to injection of botulinum neurotoxin as noted in the spasmodic dysphonia register. The spasmodic dysphonia patients were grouped into adductor spasmodic dysphonia, abductor spasmodic dysphonia and mixed spasmodic dysphonia. All patients who had associated neurological conditions, such as Meigs syndrome and vocal tremors, were excluded from the study. There were 2 Meigs syndrome patients and 13 spasmodic dysphonia patients with associated vocal tremor who were excluded from the study. The maximum phonation time of patients in all three groups was noted on their first visit to the voice clinic by the first author, prior to any injection of botulinum neurotoxin. For calculating maximum phonation time, the patients were asked to sustain phonation of vowel /e/ at a comfortable loudness level for as long as they could in one single breath, after a deep inspiration. This was repeated three times, with a gap of 40-60 seconds between each reading. The highest amongst the three was noted as the final reading. While calculating the maximum phonation time in spasmodic dysphonia patients the sustained phonation has multiple spasms and it is essential to count the maximum phonation time as the total duration of this spasmodic phonation, but only of one breath cycle.

A prospective study including 61 individuals with no vocal complaints was performed where the maximum phonation time of these individuals was noted. The age and gender of the control group were matched with those of the patients of spasmodic dysphonia. Statistical analysis was performed using the SPSS analytical tool.

Results

Out of a total of 61 spasmodic dysphonia patients, 49 (80.3 per cent) were adductor spasmodic dysphonia, 9 (14.8 per cent) were abductor spasmodic dysphonia and 3 (4.9 per cent) were mixed spasmodic dysphonia. Besides these 61 spasmodic dysphonia patients, 15 patients of spasmodic dysphonia were excluded from the study due to the presence of vocal tremors (13 patients) or Meigs syndrome (2 patients). All 61 spasmodic dysphonia patients were in the age range of 20-72 years (mean age 50 ± 13.75 years). A female predominance was noted in both adductor spasmodic dysphonia and abductor spasmodic dysphonia, with F:M ratio being 26:23 (53.1 per cent to 46.9 per cent) in adductor spasmodic dysphonia and F:M ratio being 6:3 (66.7 per cent to 33.3 per cent) in abductor spasmodic dysphonia, whereas in mixed spasmodic dysphonia, the F:M ratio was 1:2 (33.3 per cent to 66.7 per cent). An equal number of control subjects with an age range of 22-68 years $(47.26 \pm 9.74 \text{ years})$ and F:M ratio of 35:26 (57.4 per cent to 42.6 per cent) were enrolled in the study. The details of patient data in regard to age, sex, maximum phonation time of all 3 spasmodic dysphonia groups and the control group are given in Table 1.The maximum phonation time for adductor spasmodic dysphonia was 24.59 ± 7.75 seconds, for abductor spasmodic dysphonia it was 9.11 ± 3.55 seconds, and for mixed spasmodic dysphonia the maximum phonation time was 19.00 ± 4.58 seconds, as compared to the 61 controls $(16.38 \pm 3.56 \text{ seconds})$ (Fig 1). According to Dunnett's t-test, comparison of the maximum phonation time of adductor spasmodic dysphonia patients and abductor spasmodic dysphonia patients with control patients was found to be statistically significant (p < 0.001) (Table 1).

Table 1. Demographic and maximum phonation time (MPT) profile of adductor spasmodic dysphonia (ADSD), abductor spasmodic dysphonia (Mixed SD) and controls

SN	Characteristic	ADSD (n = 49)	ABSD (n = 9)	Mixed SD (n = 3)	Controls (n = 61)	Statistical significance	
1.	Mean age±SD (Range) in years	50.00 ± 12.72 (20-72)	52.78 ± 20.58 (27–77)	47.67 ± 5.86 (41–52)	47.26 ± 9.74 (22–68)	F = 0.839 NS p = 0.475 (ANOVA)	
2.	Male: Female	23 (46.9%): 26 (53.1%)	3 (33.3%): 6 (66.7%)	2 (66.7%): 1 (33.3%)	26 (42.6%): 35 (57.4%)	$\chi^2 = 1.255^{NS}$ $p = 0.740$	
3.	Mean MPT±SD (Range)	24.59 ± 7.75 (13-46)*	9.11 ± 3.55 (3-16)*	19.00 ± 4.58 (15-24)	16.38 ± 3.56 (11–35)	F = 29.376 p < 0.001	

^{*}Significant as compared to control groups using Dunnett's test; NS = not significant as compared to control groups using analysis of variance (ANOVA) test; SD = standard deviation

For differentiation of adductor spasmodic dysphonia patients from control patients, a receiver operating characteristic curve was plotted (Figure 2). Maximum phonation time had an area under the curve of 0.0850 and the cut-off value ≥ 18.50 at maximum Youden Index (J), which was 79.6 per cent sensitive and 85.2 per cent specific for differentiation of adductor spasmodic dysphonia from controls with a positive predictive value of 81.3 per cent and a negative predictive value of 83.9 per cent (Table 2).

Discussion

Diagnostic delay in the management of spasmodic dysphonia is reported to be as high as 4.34 years with an average of 3.95 physicians being consulted before a conclusive diagnosis.²⁰ This delay may be attributed to spasmodic dysphonia being a rare speech disorder occurring spontaneously with normal vocal fold appearance on a laryngoscopy, lack of awareness regarding the condition, along with unavailability of an objective test to diagnose spasmodic dysphonia.²¹ Currently the diagnosis of spasmodic dysphonia is subjective and is best elaborated by Hintze et al.,²⁰ who devised a three-tiered diagnostic approach including screening questions to expose possible spasmodic dysphonia, speech examination to identify probable spasmodic dysphonia, and videostroboscopy to exclude other laryngeal pathologies. Clinical presentation of spasmodic dysphonia is often misdiagnosed as muscle tension dysphonia and requires a high index of suspicion.²² All of these existing diagnostic challenges highlight the need for a cost-effective, objective tool to aid in the diagnosis of spasmodic dysphonia.

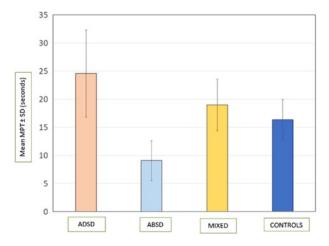
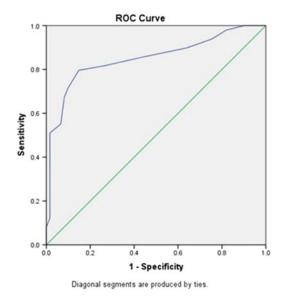


Figure 1. Bar chart showing mean with standard deviation (SD) of maximum phonation time (MPT) in adductor spasmodic dysphonia (ADSD), abductor spasmodic dysphonia (ABSD) and mixed spasmodic dysphonia (Mixed SD) patients and controls.

Maximum phonation time, an aerodynamic measure of voice, is a simple and handy clinical tool, in which the patient is asked to sustain phonation of a vowel sound (a/e/o) for the longest period in one breath. Three readings of this are taken, out of which the longest is noted. 19, 23-25 To avoid error in the calculation of maximum phonation time, clear instructions must be given to the patient to take a deep breath and phonate as long as they can. Schmidt et al.²⁶ emphasised that while taking the maximum phonation time, a demonstration of trial phonation followed by three tests with breaks of 60 seconds each should be practiced. A single rater (in this study the first author) is sufficient to get an accurate reading of the maximum phonation time.²⁷ While calculating the maximum phonation time in spasmodic dysphonia patients the sustained phonation has multiple spasms, it is essential to count the maximum phonation time as the total duration of this spasmodic phonation, but strictly of one breath. This is how we have been noting the maximum phonation time in our voice clinic over the last 15 years.

Maximum phonation time depends on various variables, including phonation volume (which itself varies with age, sex and stature), mean air-flow rate, comprehension of the task and maximal effort.²³ Normal maximum phonation time for males is 15–25 seconds and for females is 13–20 seconds.¹⁹ Ageing alters maximum phonation time²⁸ as



Line at 45 degree angle- reference line showing flip coin division

ROC line showing the area under the curve (AUC)

Figure 2. Receiver operating characteristic (ROC) curve analysis between adductor spasmodic dysphonia (ADSD) and control group, showing sensitivity and specificity.

Table 2. Receiver operating characteristic (ROC) curve analysis for derivation of cut-off value of maximum phonation time (MPT) for prediction of adductor spasmodic dysphonia (ADSD) (n = 49) vs controls (n = 61)

Area under the curve ± SE (p-value)	Youden Index (J)	Selected cut- off value at maximum Youden Index	Sensitivity	Specificity	PPV	NPV
0.850 ± 0.040 (p < 0.001)	0.648	≥ 18.50	79.6%	85.2%	81.3%	83.9%

NPV = negative predictive value; PPV = positive predictive value; SE = standard error

morphology and physiology of the larynx and adjacent organs undergo changes, such as decrease in the power of respiratory muscles, less pulmonary elasticity, and/or sarcopenia in laryngeal muscles. Blitzer *et al.*,²⁹ Patel *et al.*³⁰ and Schweinfurth *et al.*³¹ have done extensive research on spasmodic dysphonia demographics and stated that the typical ages at which symptoms first appear are 39 years old, 51 years old and 56 years old, respectively. These findings are consistent with our research, which found that spasmodic dysphonia individuals had an average age of 50.3 years.

The current study shows a greater number of female patients (53 percent in adductor spasmodic dysphonia and 66 percent in abductor spasmodic dysphonia out of a total of 61 patients studied over a span of 3 years), which is in accordance with previously conducted studies but is not high enough as projected by international studies (79.7 per cent³¹ and 80 per cent³², respectively), which may be attributed to social bias.³³ Keeping these factors of female predominance and alteration of maximum phonation time with age and gender, the control group was matched with those of the patients in our study.

- Diagnosis of spasmodic dysphonia is made subjectively by hearing the patient's voice, particularly in response to various vocal tasks, subjective videostroboscopy and exclusion of other pathologies along with a neurology opinion
- Maximum phonation time is an objective test used to assess glottic competency, which can be applied in the diagnosis of spasmodic dysphonia
- Maximum phonation time in adductor spasmodic dysphonia patients was found to be increased (> 24 seconds) and reduced in abductor spasmodic dysphonia patients (< 10 seconds) in comparison to normal subjects (16 seconds)

The diagnosis of adductor spasmodic dysphonia is mainly clinical, aided by videostroboscopy and electromyography which reveal irregular spasms of the adductor muscles during phonation.8 In abductor spasmodic dysphonia, however, the vocal fold hypo-adduction is thought to be due to spasmodic bursts in one or both of the posterior cricoarytenoid muscles, ¹⁰ which are the only abductor muscles of the larynx. Keeping in mind the hypothesis that increased activity of the adductor group of muscles in adductor spasmodic dysphonia may lead to an increased duration of the maximum phonation time and the reverse in abductor spasmodic dysphonia, the maximum phonation time was noted in all three groups of spasmodic dysphonia patients. Our study revealed that the maximum phonation time in the adductor spasmodic dysphonia group was 13-46 seconds, averaging 24.5 seconds, which was noted to be significantly higher than that of the control group where the average maximum phonation time was noted as 16.4 seconds. Patients with a maximum phonation time of 15 seconds or less in the adductor spasmodic dysphonia group (three in total) were patients with extremely severe adductor spasmodic dysphonia.

The maximum phonation time in the abductor spasmodic dysphonia group was 3-16 seconds with an average of 9.1 seconds, which was significantly lower than that of the control group. This difference is possibly due to the abductor overdrive witnessed in abductor spasmodic dysphonia. Various studies have revealed a decreased maximum phonation time in both muscle tension dysphonia and functional dysphonia.^{34–38} In muscle tension dysphonia, especially in the laryngeal isometric variety, a posterior phonatory gap may be responsible for the decreased maximum phonation time. Although there may not be a phonatory gap present in other types of muscle tension dysphonia, there is no neurological adductor overdrive described in its etiopathogenesis as in the case of adductor spasmodic dysphonia. In functional dysphonia, a phonatory gap may be a consequence of psychogenic conversion disorder and this often disappears when the patient is asked to cough.³

In mixed spasmodic dysphonia patients, the maximum phonation time was 15–24 seconds, with an average of 19 seconds, which was not statistically significant in comparison to the control group. This lack of statistical significance can be explained by the fact that patients of mixed spasmodic dysphonia have features of both adductor spasmodic dysphonia and abductor spasmodic dysphonia, which possibly nullify the effect of each other.

The limitations of our study were that it was partially a retrospective study. Furthermore, although our patients are always told in detail about the importance of prolonging the /e / to the maximum possible in one breath while checking maximum phonation time, some patients may not give their optimal maximum phonation time. However, because adductor spasmodic dysphonia showed an increase as compared to controls, this can be a factor of concern only for abductor spasmodic dysphonia. Furthermore, patients suffering from chronic obstructive pulmonary disease, severe bronchial asthma, and upper respiratory tract infections may not be ideal candidates for the test. Finally, a prospective study comparing the maximum phonation time in adductor spasmodic dysphonia, abductor spasmodic dysphonia, mixed spasmodic dysphonia, muscle tension dysphonia, functional aphonia and controls would be ideal, and we hope that our study may serve as a pilot.

We propose the use of maximum phonation time, in addition to the existing subjective vocal tests, as a cost-effective objective tool in the diagnosis of adductor spasmodic dysphonia and abductor spasmodic dysphonia. This association between maximum phonation time and spasmodic dysphonia has not been published previously.

Conclusion

Spasmodic dysphonia is currently diagnosed primarily subjectively by hearing the patient's voice, performing vocal tasks and flexible videostroboscopy. Maximum phonation time is an

objective test that can further corroborate the diagnosis of adductor spasmodic dysphonia and abductor spasmodic dysphonia in suspected cases. Average maximum phonation time was found in our study to be prolonged (> 24 seconds) in adductor spasmodic dysphonia and reduced (< 10 seconds) in abductor spasmodic dysphonia as compared to normal (control) subjects (16 seconds). Therefore, we recommend that maximum phonation time, which is a zero-cost, objective test, be added to the diagnostic armamentarium of spasmodic dysphonia.

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