- and neck surgery 12: 543-548, 2004.
- Ludlow CL, Adler CH, Berke GS, et al: Research priorities in spasmodic dysphonia. Otolaryngology head and neck surgery. Official Journal of American Academy of Otolaryngology—Head and Neck Surgery 139: 495-505, 2008.
- 3) Aronson AE, De Santo LW: Adductor spastic dysphonia: three years after recurrent laryngeal nerve resection. The Laryngoscope 93: 1-8, 1983.
- 4) Blitzer A, Brin MF, Stewart CF: Botulinum toxin management of spasmodic dysphonia (laryngeal dystonia): a 12-year experience in more than 900 patients. The Laryngoscope 108: 1435-1441, 1998.
- 5) Kobayashi T, Niimi S, Kumada M, Kosaki H, Hirose H: Botulinum toxin treatment for spasmodic dysphonia. Acta oto-laryngologica (Suppl 504): 155-157, 1993.
- 6) Nakamura K, Muta H, Watanabe Y, et al: Surgical treatment for adductor spasmodic dysphonia--efficacy of bilateral thyroarytenoid myectomy under microlaryngoscopy. Acta Oto-laryngologica 128: 1348-1353, 2008.
- 7) Isshiki N, Haji T, Yamamoto Y, Mahieu HF: Thyroplasty for adductor spasmodic dysphonia: further experiences. The Laryngoscope 111: 615-621, 2001.

- 8) 宮岡 剛, 笠原 恭, 三浦 星, 他: Clonazepam が 奏効したけいれん性発声障害の1症例. 精神医学 44: 543-545, 2002.
- 9) Thaker GK, Nguyen JA, Strauss ME, et al: Clonazepam treatment of tardive dyskinesia: a practical GABAmimetic strategy. Am J Psychiatry 147: 445-451, 1990.
- 10) Yoshimura R, Kakihara S, Soya A, et al: Effect of clonazepam treatment on antipsychotic druginduced Meige syndrome and changes in plasma levels of GABA, HVA, and MHPG during treatment. Psychiatry Clin Neurosci 55: 543-546, 2001.
- 11) Duchen LW: An electron microscopic study of the changes induced by botulinum toxin in the motor end-plates of slow and fast skeletal muscle fibres of the mouse. J Neurol Sci 14: 47-60, 1971.
- 12) 熊田 政, 村野 恵, 小林 武:【痙攣性発声障害の現 状】 Botulinum Toxin による治療。音声言語医学 42:355-361, 2001.
- 13) 小林 武:【声帯に病変のない声の異常】痙攣性発声障 害の治療 ボツリヌストキシン注射. ENTONI **105**: 12-18, 2009:
- 14) 熊田 政:【喉頭科診療における論点】痙攣性発声障害 の治療 ボツリヌストキシン注射の立場から. JOHNS 23: 1709-1712, 2007.

\* \* \*

## **ARTICLE IN PRESS**

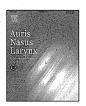
Auris Nasus Larynx xxx (2016) xxx-xxx



Contents lists available at ScienceDirect

#### Auris Nasus Larynx

journal homepage: www.elsevier.com/locate/anl



## Voice tuning with new instruments for type II thyroplasty in the treatment of adductor spasmodic dysphonia

Tetsuji Sanuki\*, Eiji Yumoto, Yutaka Toya, Yoshihiko Kumai

Department of Otolaryngology-Head & Neck Surgery, Graduate School of Medicine, Kumamoto University, Kumamoto, Japan

ARTICLE INFO

Article history: Received 30 July 2015 Accepted 23 December 2015 Available online xxx

Keywords:
Adductor spasmodic dysphonia
Type II thyroplasty
A thyroid cartilage elevator
Spacer devices

#### ABSTRACT

Objective: Adductor spasmodic dysphonia is a rare voice disorder characterized by strained and strangled voice quality with intermittent phonatory breaks and adductory vocal fold spasms. Type II thyroplasty differs from previous treatments in that this surgery does not involve any surgical intervention into the laryngeal muscle, nerve or vocal folds. Type II thyroplasty intervenes in the thyroid cartilage, which is unrelated to the lesion. This procedure, conducted with the aim of achieving lateralization of the vocal folds, requires utmost surgical caution due to the extreme delicacy of the surgical site, critically sensitive adjustment, and difficult procedures to maintain the incised cartilages at a correct position.

During surgery, the correct separation of the incised cartilage edges with voice monitoring is the most important factor determining surgical success and patient satisfaction.

*Methods:* We designed new surgical instruments: a thyroid cartilage elevator for undermining the thyroid cartilage, and spacer devices to gauge width while performing voice monitoring. These devices were designed to prevent surgical complications, and to aid in selecting the optimal size of titanium bridges while temporally maintaining a separation during voice monitoring.

Results: We designed new surgical instruments, including a thyroid cartilage elevator and spacer devices. Precise surgical procedures and performing voice tuning during surgery with the optimal separation width of the thyroid cartilage are key points for surgical success.

Conclusion: We introduce the technique of voice tuning using these surgical tools in order to achieve a better outcome with minimal surgical complications.

© 2016 Elsevier Ireland Ltd. All rights reserved.

#### 1. Introduction

"Adductor spasmodic dysphonia" (AdSD) is an idiopathic focal dystonia characterized by a strained-strangled voice quality, effortful speech production, and frequent voice breaks occurring secondary to adductor muscle spasms with consequent vocal fold hyperadduction [1]. Current treatments for SD

http://dx.doi.org/10.1016/j.anl.2015.12.015 0385-8146/© 2016 Elsevier Ireland Ltd. All rights reserved. consist primarily of botulinum toxin injections. Although injections can relieve symptoms and improve the quality of life [2,3], the approach has notable limitations. First, injections must be repeated approximately every 4 months [4]. Second, effectiveness can vary significantly with injection site and dose, as well as over time. Third, in some studies patients have reported a suboptimal voice 60% of the time [5,6]. Thus, there is a continuing need for alternative therapies.

Type II thyroplasty differs from previous treatments [7–13] in that this surgery does not involve any surgical intervention with regard to the laryngeal muscle, nerve, or vocal folds. Type II thyroplasty intervenes in the thyroid cartilage, which is

<sup>\*</sup> Corresponding author at: Department of Otolaryngology-Head & Neck Surgery, Graduate School of Medicine, Kumamoto University, Honjo, Kumamoto 860-8556, Japan. Tel.: +81 96 373 5255; fax: +81 96 373 5256.

E-mail address: otostl0319@fc.kuh.kumamoto-u.ac.jp (T. Sanuki).

T. Sanuki et al./Auris Nasus Larynx xxx (2016) xxx-xxx

2

unrelated to the lesion. This procedure, conducted with the aim of achieving lateralization of the vocal folds, requires the utmost surgical caution due to the extreme delicacy of the surgical site, critically sensitive adjustment, and difficult procedures to maintain the incised cartilages in the correct position [14]. There are reports of surgical complications, such as friable cartilages [15,16], perforation of the upper anterior commissure, and distorted vocal folds with extensive subperichondrial undermining around the anterior commissure [17].

During surgery, the correct separation of the incised cartilage edges with voice monitoring is the most important factor in determining surgical success and patient satisfaction. In 2004, Chan et al. [15] reported poor outcomes of type II thyroplasty. This was due largely to the fact that they performed the surgery under general anesthesia without voice monitoring.

We designed new surgical instruments: a thyroid cartilage elevator for undermining the thyroid cartilage, and spacer devices to gauge the width while performing voice monitoring. These devices were designed to prevent surgical complications, and to aid in selecting the optimal size of titanium bridges, while temporarily maintaining a separation during voice monitoring. In this paper, we introduce the technique of voice tuning using these surgical tools to achieve a better outcome with minimal surgical complications.

#### 2. Surgical technique and discussion

## 2.1. Essential points in surgical technique for type II thyroplasty

Under local anesthesia, a horizontal skin incision, about 3–4 cm in length, is made at a level a little lower than the midpoint of the thyroid cartilage. The median vertical strip of

the thyroid cartilage is exposed from well above the notch down to the cricothyroid membrane. The thyroid cartilage is incised at the midline, leaving the underlying soft tissue intact.

## 2.1.1. Separation between the thyroid cartilage and the inner perichondrium

Fine instruments are required for the separation and undermining of the incised thyroid cartilage. The separation is made just along the backside of the cartilage. Cartilageperichondrial separation using the new thyroid cartilage elevator (Fig. 1) is also performed laterally, from the median incision within the area to hold the titanium bridge [18]. Two titanium bridges are now used for firm and permanent fixation of the thyroid cartilage at both the upper and lower corner. The thyroid cartilage elevator has a scale on the tip for indicating the distance to undermine, usually about 2 mm from the midline at the lower portion. Similarly, the upper portion is also undermined to hold the bridge (Fig. 2). The middle part of the incised thyroid cartilage, the anterior commissure is undermined minimally, or if possible, left untouched. Undermining is not performed widely around the anterior commissure to prevent detachment of the vocal folds from the anterior commissure, resulting in the lowering of the vocal pitch [17].

## 2.1.2. Determining the optimal width of separation of the incised cartilage edges

There are spacer devices (Fig. 3) of various widths and two depths, designed to temporarily maintain the exact length of separation at the lower and upper parts of the incised thyroid cartilage, and to assist in holding the cartilage to select the optimal size titanium bridges (video). While asking the patient to produce vocal sounds, such as vowel /a/, or any words that the patient has problems pronouncing, such as "ohayo gozaimasu" (meaning: good morning), the separation width

Table 1
A summary of results.

Pt. No.	Gender	Age	Pre-Op.							3 month Post-Op.						
			F0	Flow	G	R	В	A	S	F0	Flow	G	R	В	A	S
1	F	29	245	226	1	0	0	0	1	243	131	0	0	0	0	0
2	M	39	138	141	2	0	0	0	2	166	416	1	0	0	0	1
3	M	36	118	145	2	0	0	0	2	164	147	1	0	0	0	1
4	F	29	262	175	2	0	1	0	2	264	96	1	0	0	0	1
5	F	58	232	84	2	0	0	0	2	264	88	0	0	0	0	0
6	F	24	246	136	2	0	0	0	2	264	179	1	0	0	0	0
7	M	27	117	102	3	0	0	0	3	121	100	1	0	0	0	1
8	F	76	245	309	2	0	0	0	2	254	292	0	0	0	0	0
9	F	46	209	400	3	0	1	0	2	254	100	1	0	1	0	0
10	F	47	241	70	2	1	0	0	2	248	61	0	0	0	0	0
11	F	45	218	180	2	0	0	0	2	250	140	0	0	0	0	0
12	F	34	213	222	1	0	0	0	1	210	95	0	0	0	0	0
13	F	39	252	68	2	0	0	0	2	269	62	0	0	0	0	0
14	M	43	111	101	2	0	0	0	2	148	233	1	0	1	0	0
15	F	50	253	61	2	0	0	0	2	292	122	1	0	0	0	1
16	F	28	221	71	1	0	0	0	1	253	154	0	0	0	0	0
17	F	32	232	151	1	0	0	0	1	219	130	0	0	0	0	0
18	F	44	196	74	2	0	0	0	1	187	132	0	0	0	0	0
19	F	49	199	68	3	0	0	0	2	219	119	1	0	0	0	1
20	M	47	182	93	2	1	0	0	2	175	145	0	0	0	0	0
	Ave.		206	144	2.0	0.1	0.1	0.0	1.8	223	147	0.5	0.0	0.1	0.0	0.3

Please cite this article in press as: Sanuki T, et al. Voice tuning with new instruments for type II thyroplasty in the treatment of adductor spasmodic dysphonia. Auris Nasus Larynx (2016), http://dx.doi.org/10.1016/j.anl.2015.12.015



Fig. 1. The thyroid cartilage elevator. The thyroid cartilage elevator has an angle and a scale on the tip indicating the distance to be undermined.

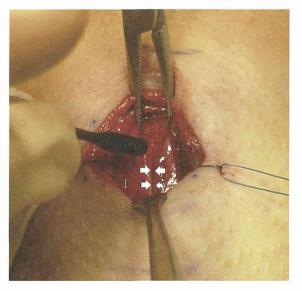
is adjusted to the optimal point where the voice can easily be produced without any strangulation sensation being experienced by the patient. Too wide a separation can make the voice breathy and weak. At the moment when the glottis on phonation is adequately widened, the patient always realizes the difference in the ease of voice production. The spacer devices may be identical, or they may be different sizes at the upper and lower portions of the thyroid cartilage, depending on voice improvement. An adequate width of separation ranges from 2 to 6 mm, most commonly 3–4 mm.

## 2.1.3. Installation of the selected titanium bridges and fixation by sutures

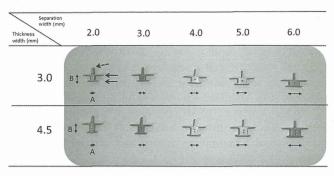
After confirming the space created under the thyroid cartilage to hold the bridge, selected bridges of the same sizes as the spacer devices are carefully bent with pliers to comply with the curvature of the thyroid cartilage, and are gently set holding the cartilage, one side after the other, with the help of an assistant surgeon. The edges of the incised cartilage should not be damaged, which would cause narrowing of the intended width and reduce the effect of the surgery. The bridges are fixed to the cartilage with four 4–0 nylon sutures, passing through the hole or holding them using a suture surrounding the bridge.

#### 2.2. The benefits of new surgical instruments

Ethical approval for this study was obtained from the Institutional Review Board of Kumamoto University Hospital.



**Fig. 2.** The thyroid cartilage elevator and curved-tip hemostatic forceps. These are used to undermine laterally from the median incision within the area to hold the titanium bridges apart, usually about 2 mm. White arrows: the incised edges of thyroid cartilage.



**Fig. 3.** Spacer devices. The devices have an assist handle (dotted-line arrow) and a short bilateral receptor (solid-line arrows) to grasp the cartilage edges, because the devices are used to temporarily maintain the correct separation while deciding on the optimal width of separation of the incised cartilage edges. The spacers have five different separation widths (A = 2.0-6.0 mm) with five different colors, the same as the titanium bridges (double-lined arrow), and are able to accommodate the different cartilage thicknesses of male and female patients (B = 3.0 and 4.5 mm).

We attempted to use the surgical instruments for type II thyroplasty in 20 cases of AdSD.

- (1) The thyroid cartilage elevator makes it easier to undermine the incised edges of the thyroid cartilage because the elevator has a flat, angled tip to fit precisely. During the cartilage-perichondrial undermining around the anterior commissure, the elevator indicates the distance to precisely undermine the cartilage perichondrium around the anterior commissure. There were no surgical complications, such as friable cartilage or lowering of the vocal pitch in this study.
- (2) While asking the patient to produce vocal sounds, such as the vowel/a/and other words, the separation width of the incised thyroid cartilage is adjusted to the optimal point where the voice can be produced easily without any strangulation sensation being experienced by the patient. However, holding the incised thyroid cartilage with a pair of curved-tip hemostatic forceps results in an unstable phonation because the surgeon holds the forceps during phonation. The spacer devices are the same size as the corresponding titanium bridges. The devices are used to temporarily maintain the correct separation while determining the optimal width of separation of the incised cartilage edges. Several sizes of the devices are tried repeatedly until patients are satisfied with their voice change during surgery. Spacers of two thicknesses were prepared to accommodate differences in male and female thyroid cartilage thickness. Spacers accommodating a cartilage thickness of 3.0 mm are generally used for female patients, while spacers accommodating a cartilage thickness of 4.5 mm are used for male patients.
- (3) Table 1 shows a summary of results. After surgery, GRBAS scale showed significant improvement in overall grade and sustain grade. There is no lowering pitch after surgery.

#### 3. Conclusions

We designed new surgical instruments, including a thyroid cartilage elevator and spacer devices. Precise surgical

## **ARTICLE IN PRESS**

T. Sanuki et al./Auris Nasus Larynx xxx (2016) xxx-xxx

procedures and performing voice tuning during surgery with the optimal separation width of the thyroid cartilage are key points for surgical success. Using these tools and techniques in the treatment of 20 AdSD patients (from 2011 to 2013) resulted in superior patient satisfaction with no complications.

#### Conflict of interest

There is no conflict of interest to be disclosed with any companies.

#### Acknowledgment

This study is partially supported by the Practical Research Project for Rare/Intractable Diseases (15ek0109006) from Japan Agency for Medical Research and Development, AMED.

#### Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at http://dx.doi.org/10.1016/j.anl. 2015.12.015.

#### References

- Cannito MP, Johnson JP. Spastic dysphonia: a continuum disorder. J Commun Disord 1981;14:215–33.
- [2] Troung DD, Rontal M, Rolnick M, Aronson AE, Mistura K. Doubleblind controlled study of botulinum toxin in adductor spasmodic dysphonia. Laryngoscope 1991;101(Pt 1):630–4.
- [3] Blitzer A, Brin MF, Stewart CF. Botulinum toxin management of spasmodic dysphonia (laryngeal dystonia): a 12-year experience in more than 900 patients. Laryngoscope 1998;108:1435–41.
- [4] Blitzer A, Brin MF, Stewart C, Aviv JE, Fahn S. Abductor laryngeal dystonia: a series treated with botulinum toxin. Laryngoscope 1992;102;163–7.

- [5] Paniello RC, Barlow J, Serna JS. Longitudinal follow-up of adductor spasmodic dysphonia patients after botulinum toxin injection: quality of life results. Laryngoscope 2008;118:564–8.
- [6] Novakovic D, Waters HH, D'Elia JB, Blitzer A. Botulinum toxin treatment of adductor spasmodic dysphonia: longitudinal functional outcomes. Laryngoscope 2011;121:606–12.
- [7] Blitzer A, Brin MF, Fahn S, Lovelace RE. Localized injections of botulinum toxin for the treatment of focal laryngeal dystonia (spastic dysphonia). Laryngoscope 1988;98:193–7.
- [8] Dedo HH. Recurrent laryngeal nerve section for spastic dysphonia. Ann Otol Rhinol Laryngol 1976;85(Pt 1):451–9.
- [9] Carpenter 3rd RJ, Henley-Cohn JL, Snyder 3rd GG. Spastic dysphonia: treatment by selective section of the recurrent laryngeal nerve. Laryngoscope 1979;89:2000–3.
- [10] Friedman M, Toriumi DM, Grybauskas VT, Applebaum EL. Implantation of a recurrent laryngeal nerve stimulator for the treatment of spastic dysphonia. Ann Otol Rhinol Laryngol 1989;98:130–4.
- [11] Woo P. Carbon dioxide laser-assisted thyroarytenoid myomectomy. Lasers Surg Med 1990;10:438–43.
- [12] Genack SH, Woo P, Colton RH, Goyette D. Partial thyroarytenoid myectomy: an animal study investigating a proposed new treatment for adductor spasmodic dysphonia. Otolaryngol Head Neck Surg 1993;108:256–64.
- [13] Weed DT, Jewett BS, Rainey C, Zealear DL, Stone RE, Ossoff RH, et al. Long-term follow-up of recurrent laryngeal nerve avulsion for the treatment of spastic dysphonia. Ann Otol Rhinol Laryngol 1996;105:592–601.
- [14] Isshiki N, Sanuki T. Surgical tips for type II thyroplasty for adductor spasmodic dysphonia: modified technique after reviewing unsatisfactory cases. Acta Otolaryngol (Stockh) 2010;130:275–80.
- [15] Chan SW, Baxter M, Oates J, Yorston A. Long-term results of type II thyroplasty for adductor spasmodic dysphonia. Laryngoscope 2004;114:1604–8.
- [16] Sanuki T, Isshiki N. Outcomes of type II thyroplasty for adductor spasmodic dysphonia: analysis of revision and unsatisfactory cases. Acta Otolaryngol 2008;1–7.
- [17] Kanazawa H, Isshiki N. Complications in Type II thyroplasty due to inadequate fixation. J Jpn Bronchoesophagol Soc 2008;59:497–505.
- [18] Isshiki N, Yamamoto I, Fukagai S. Type 2 thyroplasty for spasmodic dysphonia: fixation using a titanium bridge. Acta Otolaryngol (Stockh) 2004;124:309–12.

J

# Voice Onset Time for the Word-Initial Voiceless Consonant /t/ in Japanese Spasmodic Dysphonia—A Comparison With Normal Controls

\*,†Saori Yanagida, \*,†Noriko Nishizawa, †Kenji Mizoguchi, †Hiromitsu Hatakeyama, and †Satoshi Fukuda, \*†Sapporo, Hokkaido, Japan

**Summary: Objectives.** Voice onset time (VOT) for word-initial voiceless consonants in adductor spasmodic dysphonia (ADSD) and abductor spasmodic dysphonia (ABSD) patients were measured to determine (1) which acoustic measures differed from the controls and (2) whether acoustic measures were related to the pause or silence between the test word and the preceding word.

**Methods.** Forty-eight patients with ADSD and nine patients with ABSD, as well as 20 matched normal controls read a story in which the word "taiyo" (the sun) was repeated three times, each differentiated by the position of the word in the sentence. The target of measurement was the VOT for the word-initial voiceless consonant /t/.

**Results.** When the target syllable appeared in a sentence following a comma, or at the beginning of a sentence following a period, the ABSD patients' VOTs were significantly longer than those of the ADSD patients and controls. Abnormal prolongation of the VOTs was related to the pause or silence between the test word and the preceding word. **Conclusions.** VOTs in spasmodic dysphonia (SD) may vary according to the SD subtype or speaking conditions. VOT measurement was suggested to be a useful method for quantifying voice symptoms in SD.

Key Words: Adductor spasmodic dysphonia-Abductor spasmodic dysphonia-Acoustic analysis-Voice onset time.

#### INTRODUCTION

Spasmodic dysphonia (SD) is defined as a focal dystonia of the laryngeal muscles. 1 Criteria for SD as an independent voice disorder have not yet been established, and the current standardized assessment method is insufficient for the objective evaluation of its severity or effects. SD subtypes include adductor spasmodic dysphonia (ADSD), abductor spasmodic dysphonia (ABSD), and mixed spasmodic dysphonia.<sup>2</sup> Hyperadduction of the true vocal folds occurs during speech in ADSD, and the voice is hoarse and strained. In contrast, endoscopic observation of the vocal folds in ABSD patients during running speech has revealed that, with each breathy air release, a synchronous and untimely abduction of the true and false vocal folds occurs. Various methods have been used to characterize the severity of dysphonia and to evaluate the effects of treatment in patients with SD. These have included the voice handicap index<sup>3</sup> or voice-related quality of life,<sup>4,5</sup> and the acoustic, <sup>2,6–9</sup> aerodynamic, <sup>10–13</sup> and perceptual assessments<sup>10,14,15</sup> such as the Unified Spasmodic Dysphonia Rating Scale<sup>16</sup> or the Consensus Auditory Perceptual Evaluation of Voice.<sup>14</sup> Acoustic methods have focused on the assessment of vocal spasm severity, eg, the number of voice breaks, fundamental frequency (F<sub>0</sub>), maximum phonation time (MPT), and voice onset time (VOT).

Edgar et al<sup>2</sup> measured VOTs for three voiceless consonants /k, p, t/ in two different sentences. As a result, the VOTs for

voiceless consonants in ABSD patients were significantly longer than those in the controls. The purpose of this study was to investigate how the voice control difficulties in SD patients influence the segmental structure of Japanese speech production by means of VOT measurement with special reference to pauses or silence preceding the test word.

#### **METHODS**

#### **Participants**

Forty-eight patients with ADSD (40 women and eight men, mean age of  $29.2 \pm 9.9$  years) and nine patients with ABSD (eight women and a man, mean age of  $27.1 \pm 6.5$  years), evaluated at the Health Sciences University of Hokkaido Hospital between April 2008 and July 2013, participated as the subjects in this study. The diagnosis of SD was made by an otolaryngologist in conjunction with a speech-language-hearing therapist (ST) based on the results of a detailed clinical history, endoscopic observation of the larynx combined with videostroboscopy, and the acoustic and aerodynamic analysis of each patient's voice. Twenty normal controls (NC) without voice disorders (10 women and 10 men, mean age of  $27.0 \pm 6.8$  years) were also tested.

Criteria for inclusion in this study for all participants were (1) normal resonance, and language ability as judged by a licensed, certified, ST; (2) no diagnosis of neurological disorder such as Parkinson disease, cerebrovascular attack (CVA), or essential tremor; and (3) no history of professional singing or voice training. Other specific criteria for the participants with SD included (1) no previous treatment with recurrent laryngeal nerve surgery, (2) no previous treatment with botulinum toxin injection, (3) evidence of repetitive hyperadduction/abduction during speech on videoendoscopic examination, (4) symptomatic at the time of testing, and (5) no voice change on stimulus with facilitation techniques such as muscle relaxation and resonance focus.

0892-1997/\$36.00 © 2015 The Voice Foundation

http://dx.doi.org/10.1016/j.jvoice.2014.09.028

Accepted for publication September 25, 2014.

From the \*Department of Communication Disorders, School of Psychological Sciences, Health Sciences University of Hokkaido, Sapporo, Hokkaido, Japan; and the †Department of Otolaryngology-Head and Neck Surgery, Hokkaido University Graduate School of Medicine, Sapporo, Hokkaido, Japan.

Address correspondence and reprint requests to Saori Yanagida, Department of Communication Disorders, School of Psychological Sciences, Health Science University of Hokkaido, 2-5, Ainosato, Sapporo, Hokkaido 0028072, Japan. E-mail: s.yanagi@hoku-iryo-u.ac.jp Journal of Voice, Vol. 29, No. 4, pp. 450-454

#### Speech materials

Our diagnostic protocol consisted of several speech tasks in a natural speaking voice. The tasks included production of (1) sustained vowels/i, e, a, o, u/, (2) MPT for /a/, (3) counting from 0 to 50, (4) reading a story, and (5) repeating sentences which elicit voice symptoms associated with SD (cf., adductor or abductor sentences).

From our diagnostic protocol, we selected the reading task for analysis. Text for reading was "The North Wind and the Sun," ("Kitakaze to Taiyo" in Japanese). The word "Taiyo" (the Sun) was repeated three times, each time differentiated by its position in the sentence: (1) in a sentence without preceding punctuation, (2) in a sentence following a comma, and (3) at the beginning of a sentence following a period (Table 1). The target of the measurement was the VOT for the word-initial voiceless consonant /t/ in "Taiyo."

#### **Acoustic measures**

We performed acoustic recordings at the first medical examination. All patients were recorded in a quiet room using a digital audio recorder (Frontier TASCAM HD-P2 portable stereo audio recorder, TEAC Corp, Tokyo, Japan) and a condenser microphone (AKG C1000 S CE Harman International, Stamford, CT) under similar conditions. The microphone was maintained at a distance of 20 cm from the lips. The acoustic waveform was displayed using the *Praat* free software program, <sup>17</sup> and the pause or silence and VOTs were measured. The pause or silence was measured as the length of time between the spike of the test word and the final pulse of the preceding word (Figure 1). According to Edgar et al, <sup>2</sup> VOT for the test word was measured as the interval between the release of an oral constriction and the start of glottal pulsing.

#### Reliability

Intrajudge reliability was determined by the measurement of VOT. The same investigator rescored all measures without access to the original scores. Pearson r correlations were calcu-

lated for the two sets of measures. Correlation coefficients showed high intrameasurer reliability ranging from r = 0.924 to r = 1.000.

#### Statistical analysis

Using the statistical analysis tool SPSS Statistics 21, we performed a two-way repeated measures analysis of variance (ANOVA) with sentence type (in a sentence without preceding punctuation, in a sentence following a comma, and at the beginning of a sentence following a period) and subject type (ADSD, ABSD, and NC) as factors. A Greenhouse-Geisser correction for sphericity was used where necessary. To follow-up this interaction, a one-way repeated measures ANOVA was conducted separately for each level of sentence type and subject type using Bonferroni corrected *P* values.

#### **RESULTS**

#### Pause or silence

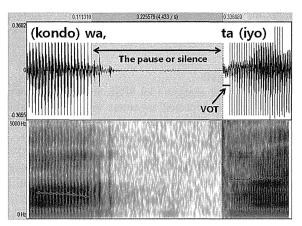
The pause or silence between the test word and the preceding word tended to be prolonged successively under conditions 1 to 3. In ADSD patients, the mean pause or silence were  $91.27 \pm 90.75$  ms under condition 1,  $256.75 \pm 162.45$  ms under condition 2, and  $894.73 \pm 376.74$  ms under condition 3. Similarly, in ABSD patients, values were  $108.78 \pm 69.18$  ms,  $239.11 \pm 125.02$  ms, and  $826.00 \pm 194.49$  ms, respectively, whereas those in the controls were  $76.05 \pm 19.25$  ms,  $254.30 \pm 207.67$  ms, and  $1014.55 \pm 301.66$  ms (Figure 2).

#### Voice onset time

In ADSD patients, the mean VOT was  $26.38 \pm 17.38$  ms under condition 1,  $34.85 \pm 23.40$  ms under condition 2, and  $45.25 \pm 29.31$  ms under condition 3. Similarly, in ABSD patients, the values were  $46.00 \pm 48.82$  ms,  $98.33 \pm 67.96$  ms, and  $134.33 \pm 103.03$  ms, respectively, whereas those in the controls were  $17.95 \pm 9.57$  ms,  $26.40 \pm 12.79$  ms, and  $25.65 \pm 10.35$  ms.

The two-way ANOVA showed a significant main effect for subject type (F(2, 74) = 28.24, P < 0.001), but no main effect

Condition	Sentences					
1. Without punctuation	Aruhi Kitakaze to <b>Taiyo</b> ga chikara-kurabe o shimashita.					
	(The North Wind and the Sun had a quarrel about which of them was the stronger.)					
2. With a comma	Kondo wa, <b>Taiyo</b> no ban ni narimashita.					
	(Then the Sun began to shine.)					
3. With a Period	Kondo wa, Taiyo no ban ni narimashita. <b>Taiyo</b> wa kumo no aida kara kao o dashite atatakana hizashi o okurimashita. Tabibito wa dandan yoi kokoromochi ni nari shimai niwa gaitou o nugisute mashita.					
	(Then the Sun began to shine. At first his beams were gentle, and in the pleasant warmth afte the bitter cold of the North Wind, the Traveler unfastened his cloak and let it hang loosely from his shoulders. The Sun's rays grew warmer and warmer. The man took off his cap and mopped his brow. At last he became so heated that he pulled off his cloak, and, to escape the blazing sunshine, threw himself down in the welcome shade of a tree by the roadside.)					

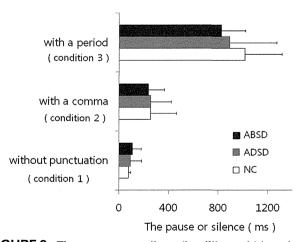


**FIGURE 1.** An example of the measures of the pause or silence and VOT under the condition 2 in controls.

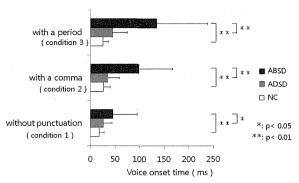
for sentence type (F(1.7, 126.3) = 26.23, P < 0.001). The interaction between the two factors was shown to be significant (F(3.4, 126.3) = 8.02, P < 0.001). To follow-up this interaction, a one-way repeated measures ANOVA using Bonferroni correction was conducted separately for each sentence type and subject type.

#### Comparison among subject groups

We compared subject types for each sentence type. For all of three sentence types, the VOTs in the ABSD patients were significantly longer than those in the ADSD patients and NC (Figure 3). Under condition 1, significant differences were observed between subject types (F(2, 74) = 5.16, P < 0.01). The VOTs in the ABSD patients were significantly longer than those in the ADSD patients (P = 0.046) and controls (P = 0.006). Significant differences were also observed between subject types under condition 2 (F(2, 74) = 20.02, P < 0.001). Again, the VOTs in the ABSD patients were significantly longer than those in the ADSD patients (P < 0.001) and controls (P < 0.001). Similarly,



**FIGURE 2.** The mean pause or silence (in milliseconds) in each patient with abductor (ABSD) and adductor spasmodic dysphonia (ADSD) and normal control (NC) subjects. The pause or silence tended to be prolonged gradually under the condition 1 to 3.



**FIGURE 3.** Comparison among subject groups. The mean voice onset time (VOT) (in milliseconds) in each patient with ABSD and ADSD and NC subjects. In all of three sentence types, the ABSD's VOTs were significantly longer than that of the ADSD and of NC.

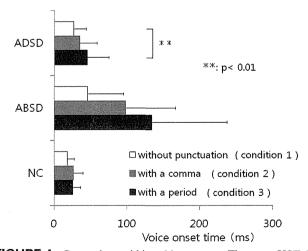
there were significant differences between subject types under condition 3 (F(2, 74) = 22.35, P < 0.001), with the VOTs in the ABSD patients were significantly longer than those in the ADSD patients (P < 0.001) and controls (P < 0.001).

#### Comparison within subject groups

Intragroup comparisons were also examined among sentence types. Significant differences among sentence type were observed for the ADSD patients (F(2, 141) = 7.53, P = 0.001). VOTs under condition 3 were significantly longer than those under condition 1. No significant differences under any conditions were observed for either the ABSD patients (F(2, 24) = 3.02, P = 0.067) or controls (F(2, 57) = 3.62, P = 0.055) (Figure 4).

#### **DISCUSSION**

In this study, one of the criteria for inclusion was the absence of any diagnosis of neurological disorders such as Parkinson disease, CVA, or essential tremor. We also excluded subjects



**FIGURE 4.** Comparison within subject groups. The mean VOT (in milliseconds) in each patient with ABSD and ADSD and NC. In ADSD, VOTs under the condition 3 were significantly longer than those of under condition 1.

with "essential voice tremor." Voice tremor is characterized by periodic fluctuations in the voice frequency or intensity at a rate between 3 and 12 Hz. <sup>18</sup> The condition is differentiated from SD, which is characterized by intermittent but nonperiodic voice stoppages or breaks. In SD, a tremorous voice may appear as abnormal phonatory behavior affecting the perception and/or differential diagnosis of ADSD from ABSD. However, the symptoms of the subjects in this study were clear cut and the differential diagnoses among ADSD, ABSD, and voice tremor could be clearly made.

Pauses are generally classified into two categories: those with breaths and those without breaths. The former are often called "physiological pauses," and latter "syntactic pauses." In Japanese speech production, breaths are a prerequisite condition at the beginning of a sentence following a period, but breaths are not necessary in sentences following a comma, or in sentences without preceding punctuation. 19 Taking these Japanese linguistic features into consideration, we applied three sets of conditions: segmentation without punctuation, segmentation with a comma, and segmentation with period. These types of segmentation reflect the semantic or grammatical distance between the words. Thus, breathing is assumed to depend on syntactic rather than physiological factors. In this study, each subject read a text at a natural speaking rate, without any order in the timing of pauses or silences. Judging perceptually, all subjects read without breaths under condition 1 (without punctuation), some of subjects read with breaths under condition 2 (with a comma), and all subjects read with breaths under condition 3 (with a period). As a result, the average duration of the pause in sentences with a period tended to be longer than that in sentences with a comma and without punctuation. These results corresponded to those of previous research.<sup>19</sup>

The purpose of this study was to demonstrate the characteristic voice symptoms in SD by measuring VOTs, and to investigate whether a pause or silence between the test word and the preceding word results in variations in the VOTs. The VOTs in the subjects with ABSD were significantly longer than those in NC and the subjects with ADSD under all three conditions. A comparison among subjects with ADSD revealed that the VOTs were significantly longer under condition 3, where a breath preceded the test word, than under condition 1, where a breath did not exist before the test word. Considering these results, prolongation of the VOTs for word-initial voiceless consonants seems to be related to the preceding pause or silence in ABSD subjects. In contrast, the VOTs in the ADSD patients were not significantly longer than those in the controls. Erickson<sup>20</sup> attempted to manipulate syntactic complexity and reported that sign expression for ADSD increased with syntactic complexity and in the voiced condition. Cannito et al<sup>21</sup> reported that for ADSD, voice quality in the voiceless condition was superior to that in the voiced condition. To test the unique influence of pause or silence between the test word and the preceding word, we would have needed to make identical phonetic environments (voiced vs voiceless, consonant vs vowels) in the words preceding the pause. Furthermore, we did not specifically control for syntactic complexity. Thus, strictly, the influence of pause time cannot be distinguished from that of phonetic environment or syntactic complexity although there is also a possibility that sample size affected our results. P values in the ABSD (P=0.067) and in the controls (P=0.055) compared with the ADSD results are very close to the level of "significance," and their nonsignificance can be explained by the fact that there were fewer subjects in both the ABSD (n=9) and control (n=20) groups versus the ADSD group (n=48). Because of the small population in the ABSD group, in particularly, the nonsignificance of the findings for the ABSD and control groups could simply represent a type II error related to differences in sample size among the groups. By combining other analysis tools, these errors may be averted.

The observed difficulty in switching between the component movements of a complex task are consistent with the behavior seen in a wide range of dystonia, including focal, action-induced dystonia, and may be related to diminished cortical movement. The larynx controls the distinction between voiced and voiceless consonants based on reciprocal activation of the posterior cricoarytenoid (PCA) and arytenoid muscles. Transient activation of the PCA muscle and restraint of the arytenoid muscle cause abduction of the vocal folds during speech in voiceless consonants. That is, the PCA muscle plays a positive role in abduction activity not only during respiration but also during the production of voiceless consonants in speech.

In the present study, conditions 2 and 3 were both accompanied by a pause which might or might not be associated with breath. In such conditions, complex cooperation between the PCA and arytenoid muscles is required or both breathing and phonation. Supposing that a disturbance in the motor subroutine<sup>23</sup> in dystonia affects the voice symptoms of SD, abnormal prolongation of the VOTs in ABSD and ADSD might be explained by distortion of the central nervous system control of the coordination of the adductor and abductor laryngeal muscles.

The respiratory center is known to be located in the medulla oblongata, which is the lowest part of the brain stem. The localization of the central pattern generator for voicing is not yet identified in humans. Katada et al<sup>24</sup> reported that repetitive electrical stimulation to the midbrain periaqueductal gray and the ventrolateral pons evokes natural sounding vocalization in cat. These findings imply that the pattern generator for spontaneous phonation exists in the brain stem in mammals. Coordination between phonation and respiration for "speech" production must be differentiated from that for the spontaneous cries of a decorticated cat. However, we can at least assume that the failure of coordination between respiratory and phonatory motor controls in ABSD subjects lead to abnormal prolongation of VOTs, particularly after the pause with breath.

The present study demonstrated that, in ABSD patients, the VOTs were prolonged under conditions in which a breath did not necessarily precede the target. As for the conditions without a breath, we cannot attribute the prolongation of VOT to the failure of respiration/phonation control. Phonetically relevant properties of speech, including VOT, are influenced by individual differences in speakers, such as speaking rate and behavioral

variations in articulation.<sup>25,26</sup> It is probable that the distinction between the semantic or grammatical distance between words might affect laryngeal adductor/abductor motor coordination in SD subjects although identical phonetic environments in the words preceding the pause or control of syntactic complexity is required to clarify this. These results suggest that VOT measurement helps the differential diagnosis of mild ABSD and ADSD.

#### CONCLUSIONS

When the target syllables appeared in a sentence following a comma, or at the beginning of a sentence following a period, the VOT for the word-initial voiceless consonant in ABSD patients were significantly longer than those in the controls. Abnormal prolongation of the VOTs was related to the pause or silence between the test word and the preceding word. VOTs in SD may vary according to the SD subtype or speaking conditions. VOT measurement of the word-initial voiceless consonant was suggested to be a useful method for quantifying voice symptoms in SD.

#### REFERENCES

- Zwirner P, Murry T, Swenson M, Woodson GE. Acoustic changes in spasmodic dysphonia after botulinum toxin injection. J Voice. 1991;5:78–84.
- Edgar JD, Spanienza CM, Bidus K, Ludlow CL. Acoustic measures of symptoms in abductor spasmodic dysphonia. J Voice. 2001;15:362–372.
- Morzaria S, Damrose EJ. A comparison of the VHI, VHI-10, V-RQOL for measuring the effect of botox therapy in adductor spasmodic dysphonia. J Voice. 2012;26:378–380.
- Hogikyan ND, Wodchis WP, Spak C, Kileny PR. Longitudinal effects of botulinum toxin injections on voice-related quality of life (V-RQOL) for patients with adductor spasmodic dysphonia. J Voice. 2001;15:576–586.
- Rubin AD, Wodchis WP, Spak C, Kileny PR, Hogikyan ND. Longitudinal effects of botox injections on voice-related quality of life (V-RQOL) for patients with adductor spasmodic dysphonia. Arch Otolaryngol Head Neck Surg. 2004;130:415–420.
- Murry T, Woodson GE. Combined-modality of adductor spasmodic dysphonia with botulinum toxin and voice therapy. J Voice. 1995;9: 460-465
- Crevier-Buchman L, Laccourreye O, Papon JF, Nurit D, Brasnu N. Adductor spasmodic dysphonia: case reports with acoustic analysis following botulinum toxin injection and acupuncture. J Voice. 1997;11: 232–237.
- Sapiena CM, Murry T, Brown WS Jr. Variations in adductor spasmodic dysphonia: acoustic evidence. J Voice. 1998;12:214–222.

- Sapoenza CM, Walton S, Murry T. Adductor spasmodic dysphonia and muscle tension dysphonia: acoustic analysis of sustained phonation and reading. J Voice. 2000;14:502–520.
- Merson RM, Ginsberg AP. Spasmodic dysphonia: abductor type. A clinical report of acoustic aerodynamic and perceptual characteristics. *Laryngo-scope*. 1979;89:129–139.
- Woo P, Colton R, Casper J, Brewer D. Analysis of spasmodic dysphonia by aerodynamic and laryngostroboscopic measurements. *J Voice*. 1992;6: 344–351.
- Cantarella G, Berlusconi A, Maraschi B, Ghio A, Barbieri S. Botulinum toxin injection and airflow stability in spasmodic dysphonia. *Otolaryngol Head Neck Surg.* 2006;134:419–423.
- Sanuki T, Yumoto E, Minoda R, Kodama N. Effects of type-II thyroplasty on adductor spasmodic dysphonia. Otolaryngol Head Neck Surg. 2010;142: 540–546.
- Braden M, Johns MM III, Klein AM, Delgaudio JM, Gliman M, Hapner ER. Assessing the effectiveness of botulinum toxin injections for adductor spasmodic dysphonia: clinician and patient perception. J Voice. 2010:24:242–249.
- Cannito MP, Woodson GE, Murry T, Bender B. Perceptual analysis of spasmodic dysphonia before and after treatment. Arch Otolaryngol Head Neck Surg. 2004;130:1393–1399.
- Stewart CF, Allen EL, Tureen P, Diamond BE, Blitzer A, Brin MF. Adductor spasmodic dysphonia: standard evaluation of symptoms and severity. J Voice. 1997;11:95–103.
- Available at: http://www.fon.hum.uva.nl/praat/download\_win.html. Accessed June 2014.
- Gamboa J, Jimenez-Jimenez F, Nieto A, et al. Acoustic voice analysis in patients with essential tremor. J Voice. 1998;12:444

  –452.
- Sugito M, Oyama G. A study of pauses and breathes in reading speech. Stud Phon Speech Commun. 1990;199–211.
- Erickson ML. Effects of voicing and syntactic complexity on sign expression in adductor spasmodic dysphonia. Am J Speech Lang Pathol. 2003;12: 416–424.
- Cannito MP, Chorna LB, Kahane JC, Dworkin JM. Influence of consonant voicing characteristics on sentence production in abductor versus adductor spasmodic dysphonia. J Voice. 2014;28:394.e13–394.e22.
- Hirose H, Ushijima T. Laryngeal control for voicing distinction in Japanese consonant production. *Phonetica*. 1978;35:1–10.
- Hamano T, Kaji R, Katayama M, Kubori T, Ikeda A, Shibasaki H, Kimura J. Abnormal contingent negative variation in writer's cramp. Clin Neurophysiol. 1999;110:508–515.
- Katada A, Nonaka S, Adachi M, et al. Functional electrical stimulation of laryngeal adductor muscle restores mobility of vocal fold and improves voice sounds in cats with unilateral laryngeal paralysis. *Neurosci Res.* 2004;50:153–159.
- Klatt DH. Voice onset time, frication, and aspiration in word-initial consonant clusters. J Speech Hear Res. 1975;18:686–706.
- Allen JS, Miller JL, DeSteno D. Individual talker difference in voice-onsettime. J Acoust Soc Am. 2003;113:544–552.

