Quantification of instability of tone production in embouchure dystonia
Quantification of instability of tone production in embouchure dystonia

André Lee a,*, 1, Shinichi Furuya a,1, Masanori Morise b, Peter Iltis a, c, Eckart Altenmüller a

a University of Music, Drama and Media Hannover, Institute for Music Physiology and Musicians’ Medicine, Emmichplatz 1, 30175 Hannover, Germany
b University of Yamanashi, Interdisciplinary Graduate School of Medicine and Engineering, 400-8510 Kofu, Japan
c Gordon College, Department of Kinesiology, 01984 Wenham, MA, USA

A R T I C L E   I N F O

Article history:
Received 13 March 2014
Received in revised form
17 June 2014
Accepted 7 August 2014

Keywords:
Movement disorders
Motor control
Musicians
Task-specificity
Frequency analysis

A B S T R A C T

Introduction: Musician’s dystonia is a task-specific loss of voluntary motor control of the fingers or the embouchure. In contrast to pianists’ dystonia, which can be objectively assessed based on movement kinematics and muscular activities, no objective quantitative measure has been established for embouchure dystonia.

Methods: We focused on acoustic signals, and investigated, whether the fluctuation of the time-varying fundamental frequency of a note can provide an objective and reliable measure of embouchure dystonia.

Results: A comparison between patients with embouchure dystonia and healthy controls found a significantly higher variability of the fundamental frequency for the patients.

Conclusion: The present findings propose a new quantification and objectivation method for embouchure dystonia.

© 2014 Elsevier Ltd. All rights reserved.

1. Introduction

Musician’s dystonia (MD) is a task-specific movement disorder that is characterized by loss of voluntary control of highly trained movements [1,2]. There are two main manifestations of MD: Hand dystonia and embouchure dystonias (ED). The first one manifests itself as an involuntary cramping of one or more fingers [2,3], whereas the latter one occurs mostly in brass instrumentalists affecting lip, tongue, facial, laryngeal or masticatory muscles as well as breathing [2,4,5]. In clinical practice an assessment tool for symptoms is useful in order to not only detect a disease, but also evaluate the course of the disease i.e. improvement or deterioration. Such measures include the subjective rating scale [6] or visual inspection [4,5]. Additionally, MD in pianists was quantitatively assessed using the scale playing analysis [7]. The standard deviation (SD) of the inter-onset intervals of a C-major scale was calculated with a higher SD signifying a more severe dystonia and vice versa. The main advantage is that the measure easily applicable and is carried out at the instrument, since due to the task specificity of MD an assessment without the instrument is likely to yield unreliable results. Furthermore a recent study could precisely identify the affected fingers in pianists with the principal component analysis and the cluster analysis [8]. In upper limb dystonia the dystonic pattern is usually visible as a flexion, or more rarely an extension of the affected finger [3]. In contrast, in embouchure dystonia not only the 12 perioral muscles of the embouchure [5] but also laryngeal or tongue muscles may be affected, making visual inspection more difficult. Therefore the diagnosis and assessment of severity often relies on the sound quality produced by the musician [3]. Usually a deterioration of the sound quality such as an unfocussed, blurred sound is the earliest symptom perceived by the musician as well as the clinician. However, subjective rating of sound quality is less reliable not only between subjects but also within one subject, making a long-term evaluation difficult. Thus an objective measure is crucially necessary for early detection of the symptom.

In order to develop such a measure for ED we aimed at assessing the effect of dystonia-induced incoordination on the fluctuation of the fundamental frequency (F0) of a sustained note. We then compared the findings of the patients with healthy brass players in order to investigate, whether these parameters may be used to objectively quantify ED. Our hypothesis was a higher instability of F0 that is detectable as a higher fluctuation in patients, as a symptom of loss of fine motor control.

2. Methods

The study was approved by the local ethics committee and written informed consent was obtained from the participants.

* Corresponding author. Tel.: +49 511 3100 552; fax: +49 511 3100 557.
E-mail addresses: eckart.altenmueller@hmtm-hannover.de, andre.lee@hmtm-hannover.de (A. Lee).
1 These authors contributed equally to the manuscript and are co-first authors.
We included 7 professional musicians with ED (patients) who reported impaired playing ability when eliciting sustained notes (age 47.1 ± 11.0 years) and 10 healthy controls (controls) (age 24.2 ± 2.7 years). One patient played the horn, four the trombone, one the tuba and one the trumpet. The healthy controls were professional musicians, of which four played the horn, five the trombone and one the tuba. Participants’ characteristics are given in Table 1. Since one parameter of interest was pitch fluctuation, each participant was asked to play 6 notes in mezzo forte (medium loudness) for 5 s with maintaining loudness or pitch as precisely as possible, and without vibrato. Because of the limited sample size we chose only one loudness range to keep the number of independent variables as low as possible. For each instrument, each of the three pitch registers (low, middle and high register) included two different notes. Since the pitch registers differ for each instrument, the trombone and horn players played G2, G3, G4 and C3, C4, C5 as low, middle, high pitches, respectively. The tuba players played G1, G2, G3 and C2, C3, C4 and the trumpet players played G2, G3, G5 and C4, C5 and B-flat 5, since our pilot study found that C6 was too high. We discarded the first 500 ms of each recording, because the attack of the note might have introduced artifacts that are not in the focus of interest. Indeed, F0 extraction became less reliable when including the note attack. Likewise we discarded the last 2 s because some participants displayed unstable tone production. Thus approximately 2.5-s duration of each note was used for further analysis. Sound was recorded with a 16Bit-48 kHz microphone in a silent room.

2.1. Feature extraction

To identify a feature that represents dystonic symptom, we first extracted time-varying information of fundamental frequency (F0) from the recorded acoustic signal using a Tandem-Straight [9]. Then we computed the standard deviation of the F0 signal during each of a short moving time-window that consists of 60 ms over the 2.5-s window (i.e., from 0.5 to 3 s), and computed the median value across all windows. The average value between two tones at each of the three pitch registers was defined as a variable that represents fluctuation of time-varying F0 signal. We did not compute the standard deviation of the whole period of each trial because in some participants we observed a component of slow oscillation in F0 signal, and its frequency differed across participants. Fig. 1 depicts a time-varying F0 signal (top panel) and spectrogram (bottom panel) of one representative patient and control during production of a G2 tone.

2.2. Statistics

For the analysis of the difference in fluctuation of F0 we performed a 2-way mixed design ANOVA with group (patient, control) and pitch (low, medium, high) as independent variables. Post-hoc tests were carried out using a t-test with Bonferroni correction (adjusted p = 0.0167). All statistical analysis was performed using R (ver.3.0.1).

We excluded age-related effects by regression analysis, which yielded no significant regression between age and each of the variables (p > 0.05).

3. Results

In all patients all pitch registers were affected albeit to different degrees. Three patients reported severest problems in the low, three in the medium and one in the high pitch register (Table 1).

Table 1

<table>
<thead>
<tr>
<th>Age (years)</th>
<th>Instrument</th>
<th>Age at dystonia onset (years)</th>
<th>Duration of dystonia (years)</th>
<th>Most affected register</th>
<th>Tongue stopping</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pat. 1</td>
<td>59</td>
<td>Horn</td>
<td>8</td>
<td>47</td>
<td>12</td>
</tr>
<tr>
<td>Pat. 2</td>
<td>48</td>
<td>Trombone</td>
<td>15</td>
<td>47</td>
<td>1</td>
</tr>
<tr>
<td>Pat. 3</td>
<td>47</td>
<td>Trombone</td>
<td>10</td>
<td>35</td>
<td>12</td>
</tr>
<tr>
<td>Pat. 4</td>
<td>29</td>
<td>Tuba</td>
<td>10</td>
<td>28</td>
<td>1</td>
</tr>
<tr>
<td>Pat. 5</td>
<td>48</td>
<td>Trombone</td>
<td>13</td>
<td>55</td>
<td>7</td>
</tr>
<tr>
<td>Pat. 6</td>
<td>40</td>
<td>Trumpet</td>
<td>14</td>
<td>39</td>
<td>1</td>
</tr>
<tr>
<td>Pat. 7</td>
<td>59</td>
<td>Trombone</td>
<td>10</td>
<td>55</td>
<td>4</td>
</tr>
<tr>
<td>Contr. 1</td>
<td>28</td>
<td>Horn</td>
<td>12</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Contr. 2</td>
<td>26</td>
<td>Trombone</td>
<td>9</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Contr. 3</td>
<td>23</td>
<td>Trombone</td>
<td>16</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Contr. 4</td>
<td>26</td>
<td>Trombone</td>
<td>15</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Contr. 5</td>
<td>22</td>
<td>Trombone</td>
<td>12</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Contr. 6</td>
<td>22</td>
<td>Horn</td>
<td>10</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Contr. 7</td>
<td>24</td>
<td>Trombone</td>
<td>9</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Contr. 8</td>
<td>25</td>
<td>Horn</td>
<td>11</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Contr. 9</td>
<td>19</td>
<td>Horn</td>
<td>10</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Contr. 10</td>
<td>27</td>
<td>Tuba</td>
<td>12</td>
<td>–</td>
<td>–</td>
</tr>
</tbody>
</table>

Pat. – Patients; Contr. – Controls.

Fig. 2 displays characteristics of F0 signals in the time (left) and frequency (right) domains at each of three pitch registers in one representative patient and control. We found a higher fluctuation of F0 in patients compared with healthy controls in all pitch registers that manifested itself as an oscillation around F0 at certain frequency peaks between about 5 and 25 Hz. Four distinct peaks were present for the low and medium pitch registers, whereas for the high pitch register one sharp peak at ca. 5 Hz and a broad peak (ca. 10–21 Hz) was visible. No such peaks were visible for control subjects.

Fig. 3A depicts the F0 variability at all participants in both patient and control groups. It was obvious that the variability was larger for almost all patients as compared with all controls. The variability of F0 revealed a threshold that segregates between the groups at ca. 0.4 Hz for the low and middle pitch registers and at ca. 0.7 Hz for the high pitch register (Fig. 3).

Fig. 3B illustrates group means of the F0 variability at the three pitch registers. Two-way ANOVA with mixed design demonstrated main effects of group (F(1,15) = 17.6, p = 0.008) and pitch (F(2,30) = 12.5, p = 0.001). There was no significant interaction effect between group and pitch (F(2,30) = 1.8, p = 0.19). Post-hoc tests identified a significant group difference at each of the low (p = 0.0004), middle (p = 0.002), and high (p = 0.013) pitch registers.

4. Discussion

We are aware that due to the small sample size we present preliminary data that should be replicated in larger samples. Therefore general conclusions must be taken cautiously. Future studies should likewise address the question, whether loudness has an influence on the precision of tone production and whether the subjective experience correlates with the objective measure. In accordance with previous reports on ED [4] not all pitch registers were affected to the same extent, although all patients reported symptoms in all registers. To our knowledge, this is the first study attempting to develop a quantification method for ED. We found a significantly higher variability of F0 irrespective of pitch register in musicians with ED, providing a quantitative and reliable measure for the instability or loss of fine motor control in this task-specific dystonia. Of particular importance is readiness of data acquisition which aids in the application of the present method for clinical evaluation. Likewise important for this task-specific disorder is the fact that assessment was performed at the instrument, where...
dystonia is triggered. Difference of F0-variability was most significant for the low and the medium pitch register, where six of the seven patients reported severest impairment. This to our understanding reflects the reliability of the measure and its capability of objectivizing the subjectively perceived impairment of playing ability.

Even though fluctuation of F0 appears to be partially rhythmic (Fig. 2), tremor was not the main complaint by the patients and was not detectible as the main feature of tone unevenness during examination. One possible explanation may be that multiple frequencies are overlapping at similar amplitude, as becomes apparent in the amplitude spectrum (Fig. 2). It could be also possible that ED has several subtypes with different symptoms.

We could successfully separate patients from controls according to the F0 variability for all pitch registers. For the low and medium pitch, a threshold of about 0.4 Hz and for the high pitch a threshold of about 0.8 Hz was found as the point where patients separated from controls.

Fig. 1. F0 signals in time (top) and frequency (bottom) domains in one representative patient (left) and control (right). Fast frequency transformation (FFT) was performed using the time-varying F0 signal of the tone G2.

Fig. 2. F0 signals in time (left) and frequency (right) domains for the three pitch registers low (top), middle (middle) and high (bottom) for patients (green) and controls (blue). The higher variability of F0 in patients becomes apparent as an oscillation around F0 in the three pitch registers. The oscillations have peak frequencies between approximately 5–25 Hz. No oscillation is visible for the controls. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)
of about 0.7 Hz was detected (Fig. 3A). Mean frequency of F0 variability was about 2.5–3 times higher for the patients compared with controls (Fig. 3B).

One interesting finding was a linear increase in mean F0-variability in the patients between the low and high pitch registers, indicating a continuous deterioration of embouchure control with increasing pitch. A hypothesis may be that with increasing motor output necessary for playing higher pitch notes, over-activation becomes increasingly detrimental leading to more severe symptoms which becomes apparent in an increase of F0-variability. Likewise for most musicians with hand dystonia severity of symptoms increases with increasing tempo, which necessitates an increase in muscular activity [10]. Additionally, mal-plasticity induced alterations in the somatosensory cortex has been found in ED [11], pointing at an impaired sensorimotor integration in ED. Therefore a pathologically higher motor output leading to increased somatosensory feedback is likely to contribute symptom severity.

In many ED-patients the attack is heavily compromised, leading to a phenomenon usually labeled as “tongue-stopper” by musicians. However, we decided to exclude the first five hundred milliseconds from our analysis firstly, because the note attack made F0 extraction with the present technique less reliable and secondly, because even in healthy brass players the clarity of attack and the timing is highly variable. Furthermore, according to our results, the subsequent maintenance of sound quality (after the attack) by adjusting the muscles contributing to sound production seems to be sensitive enough to discriminate ED-patients from healthy musicians. Furthermore, if only the attack is impaired, the acoustic analysis the way we applied would be difficult to use, because the attack itself is likely too short to assess fluctuation of F0.

In conclusion we provide a quantification method for embouchure dystonia. The advantages of this method are firstly that it can be applied in addition to the less reliable subjective rating; secondly assessment occurs at the instrument, an important prerequisite in a task specific disorder. We believe that this measure has the potential of assessing improvement or deterioration in the course of ED. However, in order to assess, whether the acoustic analysis may be potentially useful to diagnose ED, future prospective studies with bigger sample sizes and with a control condition where patients hold a tone with their voice are necessary.

Funding sources

University of music, drama and media, Hannover, Germany.

Conflict of interest

The authors declare no competing financial interests.

References