

# TREATMENT EVALUATION IN MUSICIAN'S DYSTONIA USING OBJECTIVE QUANTIFICATION TOOLS: FOLLOW-UP AFTER DIFFERENT THERAPIES

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## ABSTRACT

**Background.** Focal dystonia in pianists is a task-specific movement disorder which causes loss of pianistic skills and provokes irregularities in playing. So far, no method has been available for objective quantification of the disorder.

**Methods.** Eight professional pianists suffering from focal dystonia and eight healthy professional pianists matched by age, gender and handedness were examined, using a newly developed MIDI-based Scale Analysis Software. Subjects played 10 to 15 C Major scales on a digital piano with each hand separately and in a metronome-paced tempo. Mean standard deviations of key velocities (VEL), of tone durations (DUR), of inter-onset intervals (IOI) and of tone overlaps (OVL) of all scales were compared between the groups and between both hands of subjects. In one pianist with dystonia, follow-up examinations were performed during treatment with Botulinum Toxin-A.

**Results.** The comparison between pianists with and without dystonia revealed significantly higher values for DUR and IOI in affected hands compared to healthy reference hands. After treatment with Botulinum Toxin-A, significant improvements in performance parameters were monitored by Scale Analysis.

**Conclusions.** Scale Analysis is an effective tool for quantification of focal dystonia in pianists, allowing precise follow-up examinations during treatment and evaluation of different therapies.

## 1. INTRODUCTION

Focal dystonia in pianists – also referred to as pianist's cramp – is a task-specific movement disorder which presents itself as a loss of voluntary motor control in extensively trained movements.<sup>1, 2</sup> Involuntary flexion or extension of individual fingers fundamentally impair technical skills on the instrument, and affected pianists may suffer a dramatic impact on their pianistic careers.<sup>1, 2</sup>

In the past, the extent of focal dystonia in musicians has mainly been estimated by means of visual inspection<sup>3</sup> and rating procedures,<sup>4, 5, 6</sup> employing rating scales such as the Arm Dystonia Disability Scale<sup>7</sup> or the Tubiana and Chamagne Score<sup>8</sup>. A quantification tool for objective measurement of the extent of musician's cramps having a high precision and being independent of rating methods has not been available. Follow-up examinations after treatment of musician's dystonia were mainly accomplished by subjective comparison of videographic documents.<sup>5, 6</sup> Moreover, reliable evaluation of different therapies for musician's

cramps was not possible. Since focal dystonia is task-specific,<sup>1, 2</sup> quantification has to be performed while the patient is executing the task, e.g. a pianist has to be evaluated while playing the piano. As a consequence, for focal dystonia in pianists a MIDI-based technique has been developed to objectively measure the extent of the disorder while patients are playing the piano. The principle of the method is a scale analysis of irregularities in loudness and different timing parameters. Scales are affected in the early stages of pianist's dystonia, therefore irregularities of timing parameters and loudness in C Major scales were chosen as indicators of the extent of the disorder.

## 2. PARTICIPANTS AND METHODS

Eight pianists with focal hand dystonia and eight healthy pianists were carefully matched for age, gender and handedness. All subjects of both groups were professional pianists either graduated or in advanced stages of training. The individual duration of the disorder in the patient group was between six months and eight years ( $2.9 \pm 2.5$  years) at the time of the study. The contralateral hand was unaffected in all patients. Patients with other neurological disorders or secondary dystonias were excluded from the study. Details of both groups are given in Table 1. Informed consent was obtained from all participants prior to study participation.

	pianists with dystonia	healthy pianists
male/female (n)	6 / 2	6 / 2
mean age $\pm$ SD (years)	31.9 $\pm$ 4.8	31.4 $\pm$ 6.0
right-handed / left-handed (n)	7 / 1	7 / 1

**Table 1:** Gender, age and handedness for the matched groups of pianists with and without focal dystonia

Scales were performed on a digital piano which was connected to a computer. For recording and generating MIDI-files, a commercially available music editing software was used (Musicator Win, Version 2.12). For the test, sequences of 10 to 15 C Major scales were played over two octaves (C2-C4) in both directions, with each hand separately. Scales were played in sixteenth notes, and the tempo was standardized at 120 beats per minute for a quarter note and paced by a metronome. A newly developed MIDI-based Scale Analysis Software was used to analyze key velocities (an indirect measure of loudness), tone durations, inter-onset intervals and tone overlaps for all individual notes of the scales. Standard deviations of all four parameters were calculated for each scale. Finally, the mean

standard deviations of the four parameters of all scales of a sequence were yielded (terminology: mean standard deviation of the velocities: VEL. Mean standard deviation of tone durations: DUR. Mean standard deviation of inter-onset intervals: IOI. Mean standard deviation of tone overlaps: OVL). Scale Analysis was performed for each hand and in both directions separately. Scales containing wrong notes or other mistakes regarding the order of the C-Major-scale were entirely excluded from the analysis. A minimum of 10 correct scales was required for the analysis of a sequence.

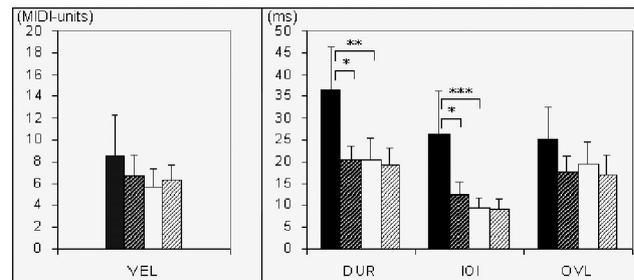
For comparison of the matched groups of dystonic and healthy pianists, the results of the four parameters of the more severely affected playing direction of the dystonic hand were focused on in each patient. Data of the affected hand and playing direction of a dystonic pianist were compared to the respective reference hand and playing direction of the matched subject. For assessment of within-group differences (affected vs. non-affected hand), Wilcoxon matched-pairs signed-rank tests were utilized. Between-group differences were analyzed with the Mann-Whitney U test.

### Follow-up after Therapy

One patient (aged 35, male, onset of pianist's cramp in the right hand eight years ago) was examined by Scale Analysis before and after treatment with Botulinum Toxin-A. After the first examination, he received a total dosage of 200 Units of Dysport® (Ipsen Limited, Berkshire, United Kingdom) which was injected into the affected muscles of the right forearm under electromyographic guidance. After a period of 147 days, the same muscles were reinjected using the same total dosage of Dysport. For monitoring of treatment response, twelve follow-up examinations were carried out using Scale Analysis. Wilcoxon matched-pairs signed-rank tests were utilized for comparison of Scale Analysis results before and after therapy.

## 3. RESULTS

Between-group analysis revealed significant differences between the affected hands of pianists suffering from focal dystonia and the reference hands of healthy subjects in the irregularities of tone durations and of inter-onset intervals (see Figure 1): DUR values were higher in dystonic hands ( $36.4 \pm 9.9$  ms) than in healthy reference hands ( $20.5 \pm 5.0$  ms;  $p < 0.01$ ). Similarly, higher values were found for IOI in affected hands ( $26.3 \pm 9.9$  ms) compared to healthy hands ( $9.5 \pm 2.1$  ms;  $p < 0.001$ ). In the VEL and OVL parameters, no significant differences were found between dystonic and reference hands. Between-group analysis between unaffected hands of pianists with dystonia and the respective reference hands of healthy pianists did not reveal significant differences in any parameter. Differences between both hands were observed only in pianists with dystonia and not in healthy subjects: within-group analysis in dystonic pianists revealed higher DUR values in affected hands ( $36.4 \pm 9.9$  ms) compared to unaffected hands ( $20.5 \pm 3.2$  ms;  $p = 0.01$ ). A similar finding was seen in IOI (affected hands:  $26.3 \pm 9.9$  ms; unaffected hands:  $12.7 \pm 2.7$  ms;  $p = 0.01$ ). No differences were found in VEL and OVL between both hands of pianists with dystonia.

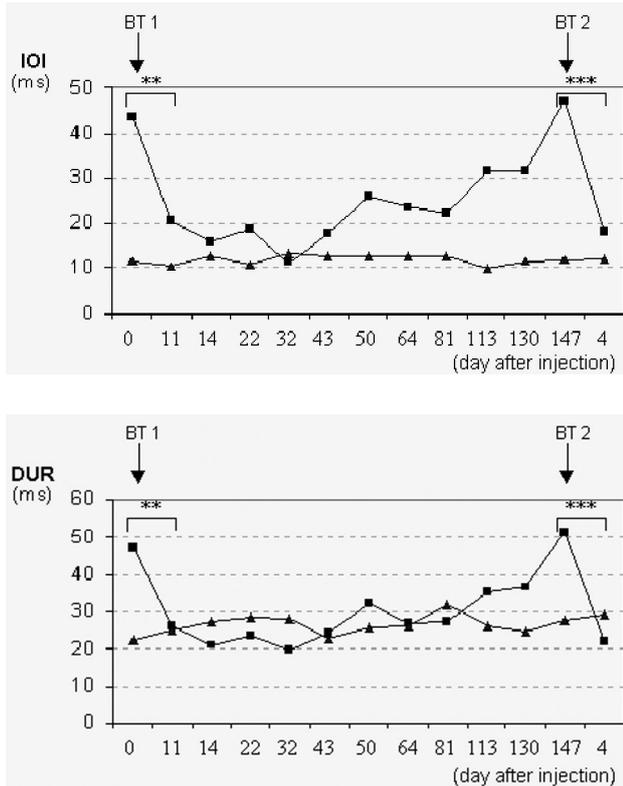


**Figure 1:** Scale Analysis in pianists with focal dystonia and in matched healthy controls. Results of affected (black bars) and unaffected (hatched bars, dark) hands of patients as well as respective reference hands of healthy pianists are shown (healthy reference for affected hands: open bars. Healthy reference for unaffected hands: hatched bars, bright). VEL: mean standard deviation of key velocities. DUR: mean standard deviation of tone durations. IOI: mean standard deviation of inter-onset intervals. OVL: mean standard deviation of tone overlaps. \* :  $p = 0.01$ ; \*\* :  $p < 0.01$ ; \*\*\* :  $p < 0.001$

One patient (Figure 2) suffering from flexion dystonia in the right hand underwent injection therapy with Botulinum Toxin-A after taking part in the study. Figure 3 displays the results of follow-up examinations after time intervals of  $13 \pm 8$  days. Significant decrease in IOI and DUR was already found in the first follow-up test on day 11 post-injection ( $p$ -values  $< 0.01$ ). After fading out of therapeutic effects of Botulinum Toxin-A, a second injection was carried out on day 147. Four days later, IOI as well as DUR were again found to be significantly lowered ( $p$ -values  $< 0.001$ ).



**Figure 2:** A pianist's right hand with moderate symptoms of focal dystonia. Overflexion of the third and fourth fingers is visible during C Major scale played in upward direction.



**Figure 3:** Scale Analysis results of a pianist with focal dystonia of the right hand during therapy with Botulinum Toxin-A (BT). Black squares: results of affected hand. Black triangles: results of unaffected hand. BT 1: date of first injection of BT. BT 2: date of reinjection of BT. IOI: mean standard deviation of inter-onset intervals. DUR: mean standard deviation of tone durations. \*\* :  $p < 0.01$ ; \*\*\* :  $p < 0.001$

#### 4. DISCUSSION

Scale Analysis has specifically been developed for precise and objective identification of subtle differences in the symptomatology of pianist's cramp such as in follow-up examinations during treatment. Taking place at the piano, the method takes into consideration the crucial fact that focal dystonia is task-specific and occurs while the patient is playing the instrument. The task of playing C Major scales reflects a critical element of piano technique which is already impaired in early stages of focal dystonia. The necessary device is inexpensive and the handling is easy. Recording time was short (approximately 10 minutes), the time of the whole procedure including analysis did not exceed 15 minutes for each subject. The comparison between pianists with dystonia and a matched group of healthy controls revealed significantly higher values for the timing parameters DUR and IOI in the dystonic hands than in healthy reference hands. This showed a clearly higher degree of irregularities in the tone durations and inter-onset intervals along the scales in affected hands. Such differences were not seen between unaffected hands of patients and reference hands of

healthy controls, indicating that irregularities in dystonic hands were not explicable by a possibly altered working behavior of pianists with dystonia.

No significant elevation was found for the VEL parameter in dystonic hands compared to reference hands, demonstrating that patients had obviously developed effective compensation mechanisms to keep the loudness largely stable. There was no significant difference between OVL values for both groups. Neither patients nor healthy pianists played with consistent overlaps. This finding might be explained by the following phenomenon: in the auditory perception, the onset of a subsequent tone may mask the detection of the ending of the preceding tone. This interaction can take place as tonal masking (in positive overlaps with a masked detection of the preceding tone after onset of the subsequent tone) and backward temporal masking (up to 50 ms before onset of the subsequent tone).<sup>9</sup> This could result in impaired perception of positive and negative overlaps. Evenness of the overlaps is perceived only to a limited extent within this range and therefore it is not an aim of practicing, even in healthy pianists.

Scale Analysis was employed for follow-up examinations during treatment with Botulinum Toxin-A in a pianist with focal dystonia. The development of the disorder was recorded over a period of 151 days. Therapeutic effects of Botulinum Toxin-A were reflected by a significant decrease of irregularities in inter-onset intervals and tone durations already measurable 11 days after the first injection and four days after the reinjection on day 147. A number of 11 follow-up examinations between both injections demonstrated the slow fading out of therapeutic effects by gradually increasing irregularities in inter-onset intervals and tone durations beginning after day 32 post-injection (Figure 3). During the period of decreased IOI and DUR values, the patient was able to perform in public again and to play technically demanding literature which had not been possible before treatment.

Among all parameters recorded by Scale Analysis, the mean standard deviation of the inter-onset intervals over the scales IOI revealed the largest differences between dystonic hands and healthy reference hands. Due to certain irregularities in the tone durations as well as in the overlaps also in healthy pianists, differences between groups were less pronounced in the DUR parameter and were not significant in the OVL parameter. For these reasons, IOI was the most sufficient parameter for reliable quantification of focal dystonia in pianists and for monitoring the development of the extent of the disorder in follow-up examinations during and after treatment.

#### 5. CONCLUSION

Scale Analysis was found to be an effective tool for quantification of focal dystonia in pianists. The handling is time-saving, inexpensive and easy. Taking place at the piano, the test is of high relevance to this task-specific disorder. IOI was the strongest parameter for describing the extent of focal dystonia and for monitoring therapeutic effects during treatment.

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