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Botulinum toxin injections in the treatment of musician's dystonia

Abstract—The authors present the results of 84 musicians with focal task-specific dystonia treated with EMG-guided botulinum toxin injections. Treatment outcome was assessed by subjective estimation of playing before and after treatment and self-rating of treatment response. Fifty-eight (69%) of the musicians experienced improvement from the injections and 30 of 84 musicians (36%) reported long-term benefit in their performance ability.

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Focal task-specific dystonia (FTSD) in musicians presents as a loss of voluntary motor control in extensively trained movements.¹ The condition affects around 1 in 200 professional musicians and often leads to the end of the musical career.² A variety of treatment options, including retraining, biofeedback, oral medication, constraint-induced therapy, and limb immobilization, may be helpful, but it is unclear if they produce sustained benefit. Botulinum toxin has been efficacious in 67 to 93% of patients with focal task-specific dystonia, but many patients eventually discontinue injections, largely because the treatment fails to meet their expectations or needs.³ This is especially so in musicians, for whom even marked improvement in the dystonia can be inadequate if the ability to play professionally is not restored.⁴

Methods. Eighty-eight consecutive musicians treated with botulinum toxin injections of a total of 226 musicians with FTSD were recruited from a musician outpatient clinic in Germany between 1995 and 2002. A retrospective chart review was performed and the patients contacted by phone using a standardized questionnaire. Patients were asked to estimate their average playing ability in percent before and during botulinum toxin treatment and to rate the cumulative treatment response using a six-step scale: worse, no improvement, mild, moderate, marked, or remission. Moderate or better improvement required that the musicians did not experience significant overshoot weakness or response fluctuations after injections, and indicated a noticeable benefit from the treatment in their performance ability. This could be either an improvement in their orchestra position, expansion of their musical repertoire, or increase in number of public performances. All

patients received lyophilized botulinum toxin A powder (Dysport, Ipsen Ltd., Berkshire, UK) using an EMG-guided technique.⁵ All dose determinations and injections were performed by one neurologist (E.A.). The group of patients who underwent at least two treatments within a period of 12 months was used to determine details of the botulinum injections. Data were statistically analyzed using the χ^2 test for outcome measures, Wilcoxon paired rank for change in playing estimation, and Student *t* test for differences in botulinum toxin dosage.

The use of botulinum toxin A injection for focal limb and embouchure dystonia is non-labeled. Informed consent was obtained from all patients prior to application.

Results. Eighty-four of 88 patients, 74 men and 10 women, 64% of them orchestra musicians, responded to the telephone survey (95%, table 1). Mean age was 45.9 years, mean duration of symptoms 10.6 years, and first botulinum toxin injection on average 6.1 years after onset. The dystonia was rated as severe in 47 musicians, moderate in 28, and mild in 9 based on the Arm Dystonia Disability Scale. Seventeen patients (20%) had a complex dystonia with spread of dystonic symptoms to other nonmusical tasks. Sixty-five musicians (77%) were diagnosed with localized hand dystonia with involvement limited to one or two fingers. Thirty-nine musicians had tried trihexyphenidyl before undergoing botulinum toxin injections, but only four were still taking the medication.

The musicians estimated their playing ability compared to the skill level prior to onset of dystonic symptoms as $53 \pm 23\%$ before and $67 \pm 23\%$ during treatment with botulinum toxin ($p < 0.005$). In the self-rating of the cumulative treatment response, 26 of 84 felt that they played worse or had no response from the injections, among them all three patients with embouchure dystonia (figure). Fifty-eight (69%) of the musicians experienced benefit from the treatment, among them 38 musicians who indicated that the treatment led to noticeable improvement in their performance ability. There was no difference in response rate between patients with localized compared to nonlocalized as well as simple compared to dystonic hand cramps. Improvement was independent of initial severity rating, sex, age at onset, and duration of symptoms prior to first injection. Ninety-eight percent of patients experienced weakness and 56% reported excessive weakness after at least one injection preceding the period of maximum improvement by 1 to 4 weeks. Twenty-four musicians continued to receive treatment at the time of the study for a mean of 36

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Table 1 Patient characteristics

Instrument group	N	Male sex	Age, y	Duration, y	Part affected	Improvement ≥ moderate (%)
All	84	74	45.9	10.6	47 R, 30 L, 4 bilateral, 3 embouchure	38 (45)
String	12	11	47	11.3	3 R, 9 L	7 (58)
Keyboard	20	17	49	9.9	15 R, 4 L, 1 bilateral	7 (35)
Guitar	25	25	45	10.8	20 R, 5 L	13 (52)
Woodwind	25	19	45	10.6	9 R, 12 L, 3 bilateral, 1 embouchure	11 (44)
Brass	2	2	38	8.0	2 Embouchure	0

months (range 9 to 76) and an average number of 11.7 injections (range 3 to 29) and six patients indicated that the dystonia had improved sufficiently after an average of 4.4 injections (range 3 to 7) and that they no longer require treatment (see the figure).

A total of 56 musicians received more than one treatment within a year (table 2). Mean duration of treatment was 23 months (range 2 to 76 months), mean interval between injections 3.8 months (range 1 to 7.7 months), average number of injections per patient 7.4 (range 2 to 29). The muscle group most frequently injected was the forearm flexors in 52 patients (93%). Average number of muscles injected per treatment was 2.5 (range 1 to 6). Average total dose per treatment was 126.9 units (range 5 to 420) at the initial visit and 112.2 units (range 3 to 1,000) at the last visit. Average dose per muscle group at last visit was 55 units for shoulder muscles (only 2 patients), 47.4 units for forearm flexors, 31.4 for forearm extensors, and 17.7 for hand muscles. No statistical difference in frequency, number of muscles, dose, and response rate was seen among the four instrument groups. No difference in average dose per treatment and muscle was seen related to sex.

Discussion. This study provides evidence that botulinum toxin offers long-term benefit for some instrumental musicians with playing-provoked, focal limb dystonia. Fifty-eight of 84 musicians (69%) responded to the treatment, similar to the initial improvement rate seen in other forms of FTSD. More

than one third of our patients reported long-term benefit and improvement in their performance ability. In a previous study, only one of 19 musicians continued treatment for longer than 2 years.⁵ Three musicians with embouchure dystonia had worsening of symptoms after the injection; treatment of these patients remains unsatisfactory.⁶

Muscles were selected by observation of playing. No booster injections were given to avoid antibody formation. In only one patient, increasing doses of botulinum toxin A failed to produce weakness after an initial response, tentatively attributed to antibody formation. This is the first study in musicians with FTSD using a preparation of botulinum toxin A (Dysport) widely used in Europe. Assuming a potency ratio of Dysport compared to Botox of 1:3, our dosing was similar to previous recommendations for botulinum toxin A for forearm muscles.⁷

A major limitation of this study is the lack of a standardized and objective assessment tool for musicians. Several attempts have been made to objectively assess outcome in writer's cramp and musician's dystonia including videotaped blind-rating of performance by musicians and neurologists.^{8,9} Recently, a MIDI-based method has been developed for rater-independent and precise quantification of focal dystonia in pianists.¹⁰ However, this technique is not applicable for other instrument groups and would have required that the participants return in between the injection cycle. Although our assessment was retrospective and subjective, the validity is supported by the fact that the musicians improved in their ability to perform and 24 musicians are continuing treatment for an average of 3 years and up to 7 years.

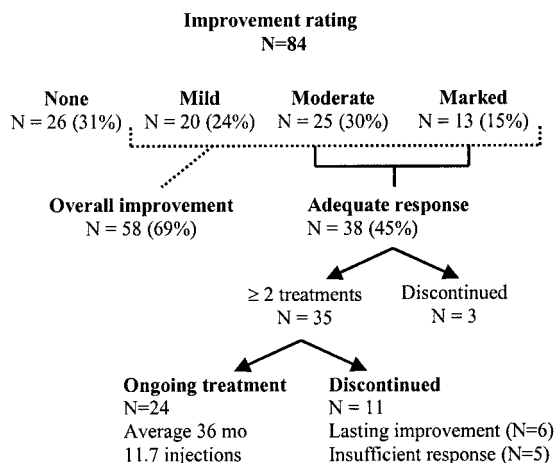


Figure. Rating of treatment response.

Table 2 Dosage of botulinum toxin A (Dysport)

Muscle group	First injection, units (range)	Last injection, units (range)	No. of patients
Shoulder	55* (40–70)	55* (40–70)	2
Forearm flexors	56.0* (10–160)	47.4* (5–250)	52
Forearm extensors	34.6* (4–100)	31.4* (4–100)	17
Hand	22.4* (4–100)	17.7* (1.5–100)	14
Total dosage	126.9 (5–420)	112.2 (3–1,000)	56

* Average dose per muscle.

References

1. Altenmüller E. Focal dystonia: advances in brain imaging and understanding of fine motor control in musicians. *Hand Clin* 2003;14:523–538.
2. Schuele S, Lederman RJ. Long-term outcome of focal dystonia in instrumental musicians. *Adv Neurol* 2004;94:261–266.
3. Karp BI. The role of botulinum toxin type A in the management of occupational dystonia and writer's cramp. In: Brin MF, Jankovic J, Hallett M, eds. *Scientific and therapeutic aspects of botulinum toxin*. Philadelphia: Lippincott Williams & Wilkins, 2000;251–258.
4. Cole RA, Cohen LG, Hallett M. Treatment of musician's cramp with botulinum toxin. *Med Probl Perf Art* 1991;6:137–143.
5. Karp BI, Cole RA, Cohen LG, Grill S, Lou JS, Hallett M. Long-term botulinum toxin treatment of focal hand dystonia. *Neurology* 1994;44:70–76.
6. Frucht SJ, Fahn S, Greene PE, et al. The natural history of embouchure dystonia. *Mov Disord* 2001;16:899–906.
7. Pullman SL, Greene P, Fahn S, Pederson SF. Approach to the treatment of limb disorders with botulinum toxin A. Experience with 187 patients. *Arch Neurol* 1996;53:617–624.
8. Cole R, Hallett M, Cohen LG. Double-blind trial of botulinum toxin for treatment of focal hand dystonia. *Mov Disord* 1995;10:466–471.
9. Priori A, Pesenti A, Cappellari A, et al. Limb immobilization for the treatment of focal occupational dystonia. *Neurology* 2001;57:405–409.
10. Jabusch HC, Vauth H, Altenmueller E. Quantification of focal dystonia in pianists using scale analysis. *Mov Disord* 2004;19:171–180.

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