Sensory mapping of lip representation in brass musicians with embouchure dystonia

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Received 26 December 2003; accepted 26 January 2004

DOI: 10.1097/01.wnr.0000120962.11505.32

Embouchure dystonia is a focal task-specific disorder involving abnormal non-coordinated movements and involuntary muscle contraction around the mouth. In professional brass players it is often so disabling that patients have to limit or give up their occupation. We examined the somatosensory homuncular representation and measured gap detection sensitivity of the lips in eight former professional musicians affected by embouchure dystonia and eight control subjects. Relative to controls, the patients' digit, and especially the thumb, representations were shifted in a lateral direction towards the lip representational zone. Patients' upper lips showed decreased sensitivity compared to their lower lips ($p < 0.01$). This asymmetry result was absent in controls. Abnormal somatosensory reorganization may contribute to the disorder. *NeuroReport* 15:815–818 © 2004 Lippincott Williams & Wilkins.

Key words: Brass players; Embouchure; Focal dystonia; Lips; Magnetoencephalography (MEG)

INTRODUCTION

Embouchure dystonia [1] is a focal dystonia characterized by uncontrollable and involuntary muscle movements of mouth, face, and jaw. This task-specific disorder is occupationally induced by extensive, repetitive, and effortful movements [2]. The disorder frequently ends the professional career of brass or woodwind players who become affected. However, few reports on embouchure dystonia provide a detailed description of the symptoms and the histories of patients [1,3]. In addition to the obvious deficits of the motoric regulation, recent studies have indicated that the functioning of the somatosensory system may also be abnormal and that this alteration may favor the development of the disorder [4–6]. Investigations in new-world monkeys have indicated that digital motor incoordination resulting from digital overuse and synchronous stimulation of the digits can be associated with an induced disorder in the representation of the digits in somatosensory cortex [7]. Also, a study demonstrated a somatotopical abnormality of digit representation in musicians suffering from focal hand dystonia by means of magnetic source imaging [8].

Like the fingertips, the lips have a high density of mechanoreceptors and they have representations in somatosensory cortex that occupy large portions of area 3b and area 1 [9]. Brass players affected by embouchure dystonia have difficulties in forming embouchure, such as a lack of lip coordination and involuntary muscle contractions of jaw movements. While there are obvious parallels between focal hand dystonia and embouchure dystonia, there are also significant differences, such as the motor regulation of digits and lips and the role of hemispheric interaction. The present study was designed to investigate if embouchure dystonia is also related to a cortical disorder or abnormality. Assuming similarity to focal hand dystonia, blurring of receptive fields would accompany embouchure dystonia. The somatosensory representation of normal subjects' lips and that of dystonic brass players' lips were compared using Magnetoencephalographic (MEG) measurements. Psychophysical testing of touch sensitivity was also performed to examine the correlation between the test results and a possibly altered somatosensory representation of the lips.

MATERIALS AND METHODS

Subjects: Eight male former professional brass players (two trumpet players, four French horn players, and two trombone players) aged 28–43 years (mean 33 ± 6) participated in this study. Although they had been diagnosed as having embouchure dystonia, none of them had received any medical treatment. None of the patients, except one with the additional symptom of subtle hyperalgesia on his right upper lip, had suffered from any other neurological disease. Despite the fact that they all lost control of their lips and some even had difficulties in controlling the position of jaw while playing, they had no symptoms when they were speaking or eating. Before being affected by the disease, they had practised intensively, playing their instruments for many years for 6 h/day on average. Their symptoms and primary areas of dystonic movements are shown in Table 1. In the case in which the jaw was diagnosed as being a dystonic area, the lips were categorized as normal. The control subjects consisted of eight male non-musicians, aged

Table 1. Patient data.

Age (years)	Instrument	Age at start of brass playing (years)	Duration of symptoms (months)	Period discontinued/play (months)	Play capability (%)	Results of neurological examination	Symptom	Sections defined as primary areas of dystonic movements
28	Trumpet	10	96	36/no	0	Normal	Lower jaw moves backward when upper lip is touched	Left and right upper lip
37	Horn	6	24	0/still play	20	Normal	Upper lip moves forward and covers mouthpiece	Left and right upper lip
28	Trombone	9	18	8/no	20	Hyperalgesia on right lip when touched	Unable to close right upper and lower lips	Right upper and lower lips
41	Trumpet	12	44	9/no	0	Normal	Focal tremor of lower jaw when forming the embouchure	Lower jaw
28	Horn	8	60	30/no	0	Normal	No clear feeling of tension in right upper lip when forming embouchure	Right upper lip
30	Horn	10	82	26/a little	70	Normal	Focal tremor of upper and lower lips	All 4 lip sites
31	Horn	16	48	16/a little	35	Normal	Left upper lip moves	Left upper lip
43	Trombone	11	60	33/a little	25	Normal	Lower jaw moves forward when mouthpiece contacts the upper lip	Both upper lips and lower jaw

23–38 years (mean 27±5), who had never received any musical training. Healthy brass players (four males and two females) were also recruited for psychophysical tests. The Ethics Commission of the Medical Faculty of the University of Münster reviewed this study. Informed consent was obtained from all subjects after the nature of the study was explained to them in accordance with the principles of the Declaration of Helsinki.

Gap detection test: All of the subjects underwent psychophysical testing in which gap detection sensitivities in their fingers and lips were examined by using cylindrical plastic domes [10]. The domes, which had gratings of equal ridge and gap widths on their surfaces (gap values: 0.35, 0.5, 0.75, 1, 1.2, 1.5, 2, and 3 mm), were placed on the lips and finger pads (digits D1–D5) in the supine position and were gently pressed. The subjects were asked whether the randomly presented gap orientation was vertical or horizontal. The sensory threshold was determined using the method of limits and the data were evaluated using nonparametric methods because lip data violated equal variance assumption.

MEG measurement of somatosensory responses: Tactile stimuli were applied to each subject using seven balloon diaphragms [11]: two diaphragms were attached to either the right or left side of upper and lower lips with adhesive tape and five were attached to ipsilateral finger pads, as in the gap detection test. The seven stimuli were presented randomly with a stimulus onset interval of 0.9–1.1 s. While the subjects laid on their stimulated side, magnetic fields were recorded from the contralateral side by a 37-channel MEG system (Magnes; 4D-Neuroimaging, San Diego, CA, USA) in a magnetically shielded and acoustically quiet room. They viewed a soundless video to distract their attention from the stimuli. After a break, the stimulated and recorded sides were switched over for the second half. The MEG data were sampled at 520.8 Hz. The MEG epochs of 748.8 ms including a baseline of 230 ms were recorded for all measurement conditions from all of the patients and control subjects, except for the right upper lip from the patient with hyperalgesia.

MEG data analysis: The corresponding somatosensory evoked fields (SEF) were selectively averaged according to the stimulated sites, and were digitally lowpass-filtered at 100 Hz. Single equivalent current dipoles (ECDs) were calculated for lips in the latency range of 26–60 ms, which were most stable. Concerning fingers, ECDs from the first major peak in 35–60 ms were calculated. The ECDs satisfying the following criteria were used for statistical evaluation: goodness of fit (GOF) > 90% and signal-to-noise ratio (based on the baseline) > 3. Further, the distance between ECDs of the upper and lower lips was calculated as differences in polar angles [12,13] of the ECDs of the respective representational locations. In addition, moments of the ECDs at peak latency of recorded signal were evaluated. The ECDs were analyzed by using ANOVA with $\alpha=0.05$ followed by Scheffé's *post hoc* test.

RESULTS

Gap detection threshold: The sensitivities of the lips and fingers are summarized in Table 2. There were no differences between the left and right fingers in each group ($p>0.444$, Wilcoxon matched-pair test) nor between groups ($p>0.99$, Kruskal–Wallis test). The lips showed lower thresholds than the fingers ($p=0.001$ in each group, Mann–Whitney test). There were no dystonia-related differences between the affected and non-afftected lips of the patients ($p=0.134$) or between the affected lips of the patients and control subjects ($p=0.138$). The patients' upper lips showed lower sensitivity than the lower lips in 10 of 16 cases; in 2 cases the results were the opposite and in 4 cases the results were equal ($z=2.59$; $p=0.010$, Wilcoxon test). No such difference was found in the healthy musicians or control subjects. The healthy musicians displayed a tendency for higher threshold than the control subjects ($p<0.06$, Mann–Whitney test).

MEG response: The waveform configurations obtained from the individual subjects' lower and upper lips with/without dystonia were similar. Representative cortical maps of both fingers and lips had the same order as that of the sensory homunculus (Fig. 1). This was confirmed by a highly significant main effect ($F(6,84)=170$, $p < 0.0001$) in three-way

Table 2. Sensory thresholds for gap detection (domes) test (mm; mean ± s.d.)

	Finger	Upper lip	Lower lip
Normal subjects	2.17 ± 0.68	1.48 ± 0.60	1.32 ± 0.42
Healthy musicians	2.15 ± 0.73	1.71 ± 0.33	1.66 ± 0.83
Patients	2.15 ± 0.72	1.94 ± 0.82*	1.33 ± 0.59
Patients' affected lips**		1.83 ± 0.83	1.75 ± 1.15
(Patients' non-affected lips)		(2.20 ± 0.84)	(1.24 ± 0.41)

*Patients' upper lips have significantly larger threshold than their lower lips ($p < 0.01$);
**affected lips include both the left and right sides.

ANOVA for group, side, and stimulation site. *Post hoc* tests revealed angle difference between the D1 s of the patients and the controls ($p < 0.009$). This bar chart focuses on the difference between the groups. The D1 to lip jump of the polar angle is smaller in the patients. A normalized measure was obtained by dividing an angle between the D1 and lip (mean of the upper and lower lips) with that of the lip value, which is the largest lateral value available. As a right upper lip ECD angle for the patient with hyperalgesia, his right lower lip ECD was applied. When these relative sizes of thumb to lip representation were submitted to two-way ANOVA with factors of side and group, there was a main effect of group ($F(1,14)=5.5$, $p < 0.04$). This confirms the observation that the relative lip to D1 difference was reduced in the patients, spanning a medial–lateral difference of 17% compared with 23% in controls (Fig. 2). The reduction was mainly due to a lateral shift of digit representation (Fig. 1). Two-way ANOVA of ECD moments with the factors of group and stimulation site showed a main effect of stimulation site ($p < 0.0001$). The ECD moments of the lips were smaller than D1, D2, and D3 ($p < 0.05$, *post hoc* test). The interaction of group and stimulation site ($p=0.016$) indicates that the ratio of finger to lip moments was larger in the controls than in the patients (Fig. 3). The moments of the same stimulation sites of the both groups were not statistically different.

DISCUSSION

Somatotopical order: The order of the cortical representations for lips and fingers of the patients and the controls is consistent with several previous reports [9,14–16]. This indicates that, in addition to the Euclidian distance and the orthogonal direction such as superior–inferior or medial–lateral, the polar angle is effective in examining somatotopical organization.

Interpretations of the reduced distance between D1 and lip: Based on the assumption that there were symptomatic analogies between focal hand dystonia and embouchure dystonia, we hypothesized that patients' homuncular organization might be distorted in the lip and face region compared with that of non-dystonic control subjects. As a result, two possibilities, an altered hand and mouth relationship and a smaller lip representation (decreased dipole moments), were explored in terms of relevance to the smaller distance of (D1–lips) representation found in the patients. Compared to the controls, the ANOVA results of the ECD moments did not support the possibility of a smaller lip representation in embouchure dystonia. However, an altered relationship between the hand and mouth representations in somatosensory cortex seems to be related with this kind of focal dystonia.

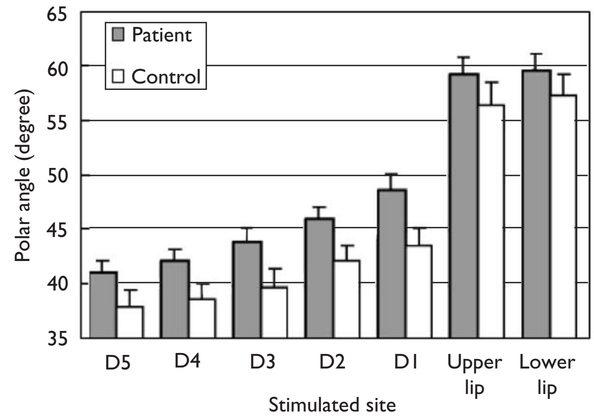

Fig. 1. Averaged polar angle of the five digits (D1–D5) and lips for both dystonic patients and controls. Error bars indicate s.e.

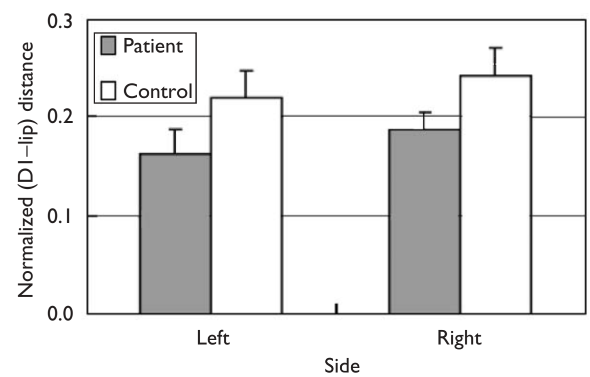

Fig. 2. Difference in polar angle between the representation of D1 and the lip was divided by the total homuncular size (measured as the polar angle of the lip in order to obtain a relative measure of homuncular size).

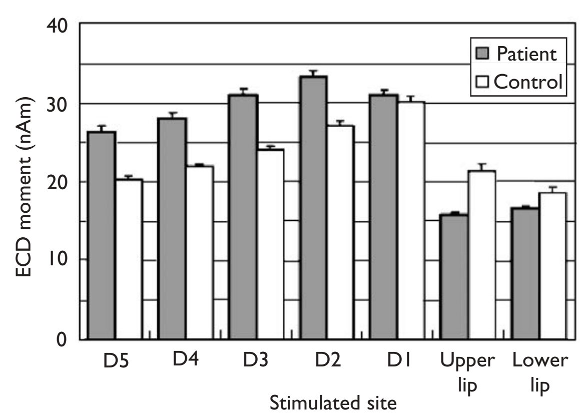

Fig. 3. Averaged ECD moment of the five digits (D1–D5) and lips for both dystonic patients and controls.

There are no distinct direct horizontal intracortical connections between representations of hand and those of lips and muzzles in monkeys [17,18]. A recent study shows that neurons located in the limb-amputated monkeys' cortical hand area 3b and in the ventro-posterior nucleus of the thalamus (the somatosensory part of the thalamus) respond to stimulation of the face [19]. This finding indicates that plasticity at sub-cortical levels occurs, and that it can change the cortical sensory maps. However, such a subcortical reorganization is not observed in a study of use-dependent plasticity in adult owl monkeys [20]. In contrast, there are cortico-cortical connections in area 3b neurons linking the hand representation to the lower jaw/neck representation [17]. These connections could cause limited cortical plasticity as a result of hard training, which might contribute to the smaller D1–lips distance representation found in the present study. Another study shows that there is an overlap of the representation of the chin and cheek and the representation of the digits in cortical area 3b of normal macaque monkeys [21]. This fact can be deduced from the evidence that monkeys, like humans, often stimulate hand and mouth synchronously. In brass players, this stimulation is more prominent for successive cooperative movements of mouth and fingers. This simultaneous stimulation could give rise to an altered hand and mouth relationship in somatosensory cortex, which might result in favoring dystonic development. The homuncular abnormality in the experimental subjects could be a correlate of the brass playing, of the dystonia or of both.

Asymmetrical gap detection threshold of embouchure dystonic patients' lips: Digital fusion even in the hemisphere opposite to the non-dystonic hand in several patients was reported by Elbert *et al.* [8]. A recent study on patients with writer's cramp has shown that both dominantly and non-dominantly affected hands have notable features of increased gap detection threshold [22]. As suggested by these findings, whether the dystonia was located in the patient's left side or right side of their lips, there was no difference in the observed sensitivities. However, our patients showed a decreased gap detection sensitivity of the upper lip compared with that of the lower lip. The upper lip of a brass player plays a major role in vibration and production of rich sounds, while the lower lip only supports the upper lip [23]. It is also quite common for musicians playing a high register to position the lower jaw slightly behind the upper jaw [24]. These techniques used by professional performers possibly result in their upper lips being highly susceptible to intense mouthpiece pressure and compression. Because the healthy musicians in the present study showed no sensitivity asymmetry, it is reasonable to infer that there is probably a close relationship between decreased sensitivity of the upper lip and occurrence of embouchure dystonia. Thus, it is still not known whether the decreased sensitivity of the upper lip is a consequence of subtle sensory nerve compression damage due to a compensatory increase in mouthpiece pressure in order to overcome the dystonic movement and to fixate the lips. Moreover, it might also be that a decreased sensitivity of the upper lip is catalyzing the development of maladaptive plasticity in the somatosensory representations.

CONCLUSION

As far as we know, this is the first study in which the functional cortical organization was investigated in musicians with embouchure dystonia. Like the shorter distance between digit representations that has been reported in focal hand dystonia, a decreased distance between lip and hand representations was observed. The increased sensory threshold of the upper lip found in the patients demonstrates that there is a positive correlation between repetitive movements of the upper lip, which is constantly pressed to a mouthpiece, and the occurrence of uncontrollable contraction of muscles around the lips.

REFERENCES

1. Frucht SJ, Fahn S, Greene PE, O'Brien C, Gelb M *et al*. The natural history of embouchure dystonia. *Mov Disord* **16**, 899–906 (2001).
2. Lim VK, Altenmüller E and Bradshaw JL. Focal dystonia: current theories. *Hum Mov Sci* **20**, 875–914 (2001).
3. Frucht SJ, Fahn S and Ford B. French horn embouchure dystonia. *Mov Disord* **14**, 171–173 (1999).
4. Leis AA, Dimitrijevic MR, Delapasse JS and Sharkey PC. Modification of cervical dystonia by selective sensory stimulation. *J Neurol Sci* **110**, 79–89 (1992).
5. Tempel LW and Perlmutter JS. Abnormal cortical responses in patients with writer's cramp. *Neurology* **43**, 2252–2257 (1993).
6. Hallett M. The neurophysiology of dystonia. *Arch Neurol* **55**, 601–603 (1998).
7. Byl N, Merzenich M and Jenkins W. A primate genesis model of focal dystonia and repetitive strain injury. *Neurology* **47**, 508–520 (1996).
8. Elbert T, Candia V, Altenmüller E, Rau H, Sterr A *et al*. Alternation of digital representations in somatosensory cortex in focal hand dystonia. *Neuroreport* **9**, 3571–3575 (1998).
9. Penfield W and Rasmussen T. *The Cerebral Cortex of Man: A Clinical Study of Localization of Function*. New York: Macmillan, 1950.
10. Van Boven RW and Johnson KO. The limit of tactile spatial resolution in humans: grating orientation discrimination at the lip, tongue, and finger. *Neurology* **44**, 2361–2366 (1994).
11. Mertens M and Lütkenhöner B. Efficient neuromagnetic determination of landmarks in the somatosensory cortex. *Clin Neurophysiol* **111**, 1478–1487 (2000).
12. Rockstroh B, Vanni S, Elbert T and Hari R. Extensive somatosensory stimulation alters somatosensory evoked fields. In: Aine C, Okada Y, Stroink G, Swithenby S, Wood C (eds). *Advances in Biomagnetism Research: Biomag96*. New York: Springer-Verlag; 2000, pp. 848–851.
13. Braun C, Haug M, Wiech K, Birbaumer N, Elbert T *et al*. Functional organization of primary somatosensory cortex depends on the focus of attention. *Neuroimage* **17**, 1451–1458 (2002).
14. Hari R, Karhu J, Hämäläinen M, Knuutila J, Salonen O *et al*. Functional organization of the human first and second somatosensory cortices: a neuromagnetic study. *Eur J Neurosci* **5**, 724–734 (1993).
15. Nakamura A, Yamada T, Goto A, Kato T, Ito K *et al*. Somatosensory homunculus as drawn by MEG. *Neuroimage* **7**, 377–386 (1998).
16. Stippich C, Hofmann R, Kapfer D, Hempel E, Heiland S *et al*. Somatotopic mapping of the human primary somatosensory cortex by fully automated tactile stimulation using functional magnetic resonance imaging. *Neurosci Lett* **277**, 25–28 (1999).
17. Manger PR, Woods TM, Munoz A and Jones EG. Hand/face border as a limiting boundary in the body representation in monkey somatosensory cortex. *J Neurosci* **17**, 6338–6351 (1997).
18. Fang PC, Jain N and Kaas JH. Few intrinsic connections cross the hand-face border of area 3b of New World monkeys. *J Comp Neurol* **454**, 310–319 (2002).
19. Florence SL, Hackett TA and Strata F. Thalamic and cortical contributions to neural plasticity after limb amputation. *J Neurophysiol* **83**, 3154–3159 (2000).
20. Wang X, Merzenich MM, Sameshima K and Jenkins WM. Remodelling of hand representation in adult cortex determined by timing of tactile stimulation. *Nature* **378**, 71–75 (1995).
21. Calford M. Limits on short-term plasticity in somatosensory cortex. In: Rowe MJ and Iwamura Y (eds). *Somatosensory Processing: From Single Neuron to Brain Imaging*. New York: Harwood Academic Publishers; 2001, pp. 153–166.
22. Sanger TD, Tarsy D and Pascual-Leone A. Abnormalities of spatial and temporal sensory discrimination in writer's cramp. *Mov Disord* **16**, 94–99 (2001).
23. Henderson HW. An experimental study of trumpet embouchure. *J Acoust Soc Am* **13**, 58–64 (1942).
24. Porter M. *The Embouchure*. London: Boosey and Hawkes, 1967.

Acknowledgements: The authors would like to thank Dr. Bernhard Ross, Andreas Wollbrink, and Karin Berning for technical help. This work was supported in part by the Ministry of Education, Culture, Sports, Science and Technology of Japan, Grant-in-Aid for Young Scientists (B), 13780627, the Deutsche Forschungsgemeinschaft (Pa 392/6-3).