Oral splint ameliorates tic symptoms in patients with tourette syndrome

Murakami, Jumpei ; Tachibana, Yoshihisa ; Akiyama, Shigehisa ; Kato, Takafumi ; Taniguchi, Aya ; Nakajima, Yoshiaki ; Shimoda, Mao ; Wake,…

(Citation)
Movement Disorders,34(10):1577-1578

(Issue Date)
2019-10

(Resource Type)
journal article

(Version)
Version of Record

Oral Splint Ameliorates Tic Symptoms in Patients With Tourette Syndrome

Tourette syndrome (TS) is a neurodevelopmental and -psychiatric disorder characterized by chronic multiple motor and phonic tics.[1] Many TS patients also complain of premonitory urges and often have comorbidities, such as attention-deficit/hyperactivity disorder, obsessive–compulsive disorder, and anxiety disorder.[2] Currently, behavioral, pharmacological, and surgical interventions are applied to TS patients.

The present study aimed to investigate whether an oral splint (Fig. 1A), commonly used for dental treatments of temporomandibular joint disorders, clenching, and bruxism, might be therapeutically effective to ameliorate TS-related tics. To this end, we examined tic scores in 22 TS patients (Supporting Information) using the Tic Symptom Self-Report, which consists of motor and phonic tic scores (0–60; higher scores indicating severer states).[3] Mean age at TS onset was 5.9 years, and mean age at the first hospital visit was 17.2 years. Mean motor and phonic tic scores before splint application were 15.3 and 15.1, respectively. Individual scores immediately decreased to 11.0 and 8.2 at the first hospital visit while wearing the splint; average reduction rates were 30% and 43%, respectively (Fig. 1B). The accompanying videos clearly demonstrate the immediate effects on the motor tics of 2 patients: Both oral and ocular tics were improved. Sixteen (72.7%) of the 22 patients exhibited improvements in both motor and phonic tics: 10 of 14 children (<20 years of age) and 6 of 8 adults experienced positive dual effects. Overall, motor and phonic tic scores were significantly improved (Fig. 1B; Wilcoxon signed-rank test: $P = 0.025$ and $P < 0.001$, respectively), and the effects were long lasting (>100 days of treatment; Supporting Information). The video-based analysis showed the effectiveness in motor tics in the head/face/neck and arm/hand/chest body parts (Supporting Information). The effects of the splint on motor tics were well correlated with those on phonic tic (Fig. 1C; Spearman's correlation coefficient: $r_s = 0.860$; $P < 0.001$). Both age at the first hospital visit and age at tic onset were critical: They were negatively correlated with long-term improvement (>100 days) of motor tic scores (Fig. 1D; Spearman's correlation coefficient: $r_s = -0.635$ and $r_s = -0.594$; $P = 0.015$ and $P = 0.025$, respectively).

Our results showed that biting this simple device successfully and immediately produced therapeutic effects on the motor and phonic tics in two-thirds of TS patients. We would like to suggest that the application of oral splint is worth trying to treat TS patients, especially younger patients. The oral splint might exert a placebo effect, but should serve as a sensory trick that ameliorates symptoms in tics[4] as well as in dystonia. Interestingly, the effectiveness of the sensory trick in cervical dystonia is associated with neural processing of proprioception.[5] The oral splint would modulate proprioceptive signals from jaw-closing muscle spindles, which are conveyed to the insular cortex[6]; the abnormal insular hyperactivity has also been reported in TS patients.[7] Further studies are needed to reconfirm the therapeutic benefit of the oral splint and elucidate its underlying mechanism.

In conclusion, an oral splint application can be therapeutically effective to ameliorate TS-related tics.

Legend to the Video

Video 1. The first and the second video segments show TS-related tics in 2 patients (see Supporting Information, No. 8 and No. 19). In each patient, the TS-related tics before the application of an oral splint are displayed on the left of screen, whereas the amelioration of tics during the splint application is shown on the right.

Jumpei Murakami, DDS, PhD,[1*]
Yoshihisa Tachibana, DDS, PhD,[2*]
Shigehisa Akiyama, DDS, PhD,[1] Takafumi Kato, DDS, PhD,[3]
Aya Taniguchi, DDS,[1] Yoshiaki Nakajima, DDS,[1]
Mao Shimoda, DDS,[1] Hiroaki Wake, MD, PhD,[2]
Yukiko Kano, MD, PhD,[4] Masahiko Takada, DDS, PhD,[5]
Atsushi Nambu, MD, PhD,[6] and
Atsushi Yoshida, DDS, PhD[7*]

[1]*Division of Special Care Dentistry, Osaka University Dental Hospital, Suita, Osaka, Japan*
[2]*Division of System Neuroscience, Kobe University Graduate School of Medicine, Kobe, Hyogo, Japan*
[3]*Department of Oral Physiology, Osaka University Graduate School of Dentistry, Suita, Osaka, Japan*

***Correspondence to:** Dr. Jumpei Murakami, Division of Special Care Dentistry, Osaka University Dental Hospital, 1-8 Yamadaoka, Suita, Osaka 565-0871, Japan; E-mail: jumpei@dent.osaka-u.ac.jp; or Dr. Yoshihisa Tachibana, Division of System Neuroscience, Kobe University Graduate School of Medicine, 7-5-1 Kusunoki, Chuo, Kobe, Hyogo, 650-0017, Japan; E-mail: yoshi@med.kobe-u.ac.jp; or Dr. Atsushi Yoshida, Department of Oral Anatomy and Neurobiology, Osaka University Graduate School of Dentistry, 1-8 Yamadaoka, Suita, Osaka 565-0871, Japan; E-mail: yoshida@dent.osaka-u.ac.jp

Drs. Murakami and Tachibana contributed equally to this work.

Funding agencies: Research Grant from Japanese Society for Disability and Oral Health (to J.M.), Grant-in-Aid for Scientific Research (18K06852 to Y.T., 26250009 to A.N., and 18K19641 to A.Y.) from the JSPS, and Grant-in-Aid for Scientific Research on Innovative Areas ("Non-linear Neuro-oscillology", 18H04946 to Y.T. and 15H05873 to A.N.) from the MEXT of Japan.

Relevant conflicts of interest/financial disclosures: Nothing to report.

Full financial disclosures and author roles may be found in the online version of this article.

Received: 11 April 2019; **Revised:** 16 July 2019; **Accepted:** 17 July 2019

Published online 00 Month 2019 in Wiley Online Library (wileyonlinelibrary.com). DOI: 10.1002/mds.27819

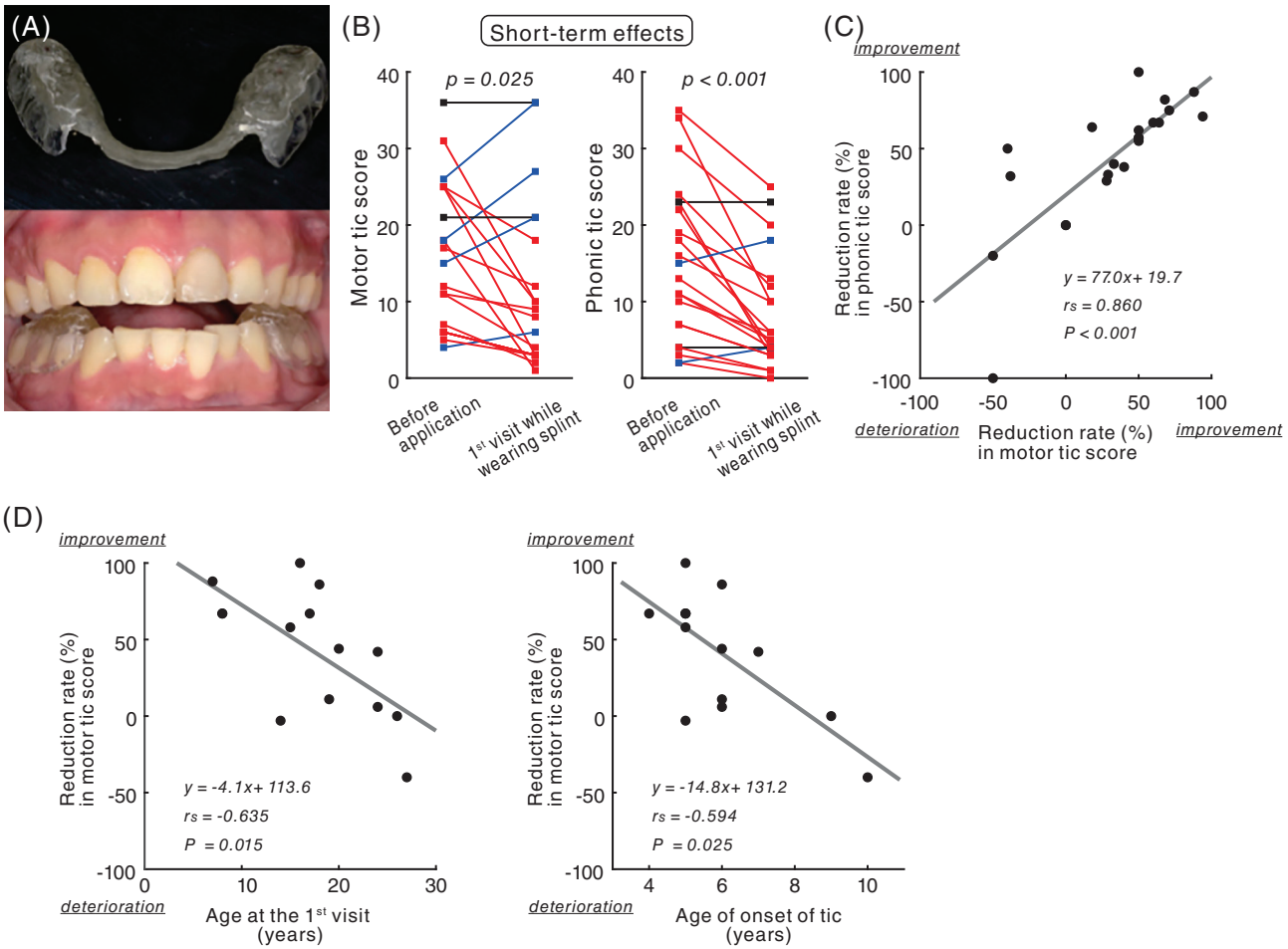

FIG. 1. Beneficial effects of oral splint on motor and phonic tics. (A) A custom-made oral splint. The splint applied to the bilateral molar teeth can increase the occlusal vertical dimension. (B) Short-term effects of the oral splint on motor (left) and phonic (right) tic scores. Using Tic Symptom Self-Report, the post-treatment effects were evaluated by the patients during a week before the first hospital visit while wearing the splint. The red lines indicate patients whose tic scores decreased while wearing the splint, the blue lines indicate patients whose scores increased, and the black lines indicate patients whose scores were unchanged. (C) Positive correlation between short-term improvements in phonic and motor tic scores. (D) Negative correlations of long-term improvements (>100 days of treatment) in motor tic scores with age at the first hospital visit (left) and with age of tic onset (right).

[4]*Department of Child Neuropsychiatry, Graduate School of Medicine, University of Tokyo, Bunkyo-ku, Tokyo, Japan*
[5]*Systems Neuroscience Section, Primate Research Institute, Kyoto University, Inuyama, Aichi, Japan*
[6]*Division of System Neurophysiology, National Institute for Physiological Sciences, and Department of Physiological Sciences, SOKENDAI (The Graduate University for Advanced Studies), Okazaki, Aichi, Japan*
[7]*Department of Oral Anatomy and Neurobiology, Osaka University Graduate School of Dentistry, Suita, Osaka, Japan*

References

1. Robertson MM. Tourette syndrome, associated conditions and the complexities of treatment. Brain 2000;123(Pt 3):425–462.
2. Leckman JF, Walker DE, Cohen DJ. Premonitory urges in Tourette's syndrome. Am J Psychiatry 1993;150:98–102.
3. Scahill L, Leckman JF, Schultz RT, et al. A placebo-controlled trial of risperidone in Tourette syndrome. Neurology 2003;60:1130–1135.
4. Wojcieszek JM, Lang AE. Gestes antagonistes in the suppression of tics: "tricks for tics". Mov Disord 1995;10:226–228.
5. Brugger F, Peters A, Georgiev D, et al. Sensory trick efficacy in cervical dystonia is linked to processing of neck proprioception. Parkinsonism Relat Disord 2019;61:50–56.
6. Sato F, Uemura Y, Kanno C, et al. Thalamo-insular pathway conveying orofacial muscle proprioception in the rat. Neuroscience 2017;365:158–178.
7. Lerner A, Bagic A, Boudreau EA, et al. Neuroimaging of neuronal circuits involved in tic generation in patients with Tourette syndrome. Neurology 2007;68:1979–1987.

Supporting Data

Additional Supporting Information may be found in the online version of this article at the publisher's web-site.

SGML and CITI Use Only
DO NOT PRINT

Author Roles

1. Research Project: A. Conception, B. Organization, C. Execution; 2. Statistical Analysis: A. Design, B. Execution, C. Review and Critique; 3. Manuscript Preparation: A. Writing the First Draft, B. Review and Critique.

J.M.: 1A, 1B, 1C, 2A, 2B, 3A
Y.T.: 1A, 1B, 2B, 2C, 3A, 3B
S.A.: 1B, 3B
T.K.: 1A, 2A, 2C, 3B
A.T.: 1C, 3B
Y.N.: 1C, 3B
M.S.: 1C, 3B
H.W.: 2C, 3B
Y.K.: 2C, 3B
M.T.: 2C, 3B
A.N.: 2C, 3B
A.Y.:1A, 1B, 2A, 2C; 3A, 3B

Financial Disclosures

Nothing to report.