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Case Report

Effects of orthopedic treatment using temporomandibular joint balancing appliance (TBA) at improving the symptoms of tic/Tourette syndrome: case report

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ABSTRACT

Tic disorder is characterized as sudden, non-rhythmic, involuntary and repetitive movement or vocalization, and its onset occurs mainly in childhood or adolescence. The development of medication has been limited because of a lack of understanding of tic mechanisms, and behavioral treatment is recommended as a first-line treatment. In this case report, we observed three cases in which tic disorder or Tourette syndrome was treated with intraoral orthopedic treatment (FCST) combined with acupuncture and an herbal formula and was assessed using the Yale Global Tic Severity Scale (YGTSS), a visual analog scale (VAS) and video analysis. The symptoms were reduced in 30 days and remained at the reduced level until 240 or 300 days after the first day of treatment. Most of the symptoms were disappeared after 240–300 days. Therefore, we could conclude that orthopedic treatment combined with Korean medicine may be applied for tic disorder in accordance with the efficacy and low adverse event rate, and we also suggests a large-scale clinical report to provide concrete evidence before the global application.

1. Introduction

Tics are defined as sudden, non-rhythmic, involuntary and repetitive movements or vocalizations.[1] Tourette syndrome (TS) is a neurodevelopmental disorder with onset mainly in childhood, and it is characterized by repetitive motor and vocal tics.[2] Genetic factors, environmental factors or neurodevelopmental factors which occurs failure of frontal-subcortical circuit maturation are considered factors related to tic disorder; however, detailed mechanisms of tic generation remain unknown.[3]

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Only two drugs, haloperidol (1967) and pimozide (1983), have been approved by the FDA because the mechanism of tics is not clear, and the first-line treatment is considered to be education or behavioral treatment.[3] Recently, fluphenazine,[4] risperidone[5] and aripiprazole[6] have been used to reduce the symptoms of TS. In addition to these conventional medications, deep brain stimulation[7] and herbal medicines[8] are listed as adjuvant treatments for tic disorder.

The functional cerebrospinal technique (FCST) is a newly developed technique including diagnosis and treatment techniques for balance of the temporomandibular joint (TMJ), developed by Lee based on applied kinesiology.[9] Recently, several cases about Torticollis[10,11] or alignment of curved and rotated cervical vertebrae[12] have been reported. Yin et al[13] and Yin et al[14] examined the mechanisms of FCST and concluded that it improves brain function by aligning the imbalanced TMJ, spinal cord or skull; however, there has not yet been any evidence proving this hypothesis.

In this case report, we aimed to report the efficacy of a combined treatment of orthopedic skill and traditional medicine, including acupuncture and herbal medicine, which had fewer adverse events than current medications, for tic disorder as to address the need for an in-depth study of the effects on the whole body system of orthopedic treatment at the TMJ.

2. Methods

2.1. Treatment

2.1.1. FCST therapy

For each patient, FCST treatment was applied after FCST applicability was identified using a manual test. The brief procedures of the FCST operation were as follows.

First, the deviation between the left and right TMJs was measured using a manual test[15] to confirm the balance of the TMJ by checking the locking with pain in the cervical area caused by high tension of the muscle and using test films with equal thickness (0.8 mm and 0.05 mm, Jin biotech, Cheonan, Korea).

A customized oral orthopedic device was manufactured using dental putty (base and catalyst, exafine putty type, GC Corporation, Tokyo, Japan) and applied in the patients. To introduce the manufacturing process of the customized oral orthopedic device briefly, we set the equivalence of the TMJ by inserting test films between the upper and lower molars. The equivalence was confirmed with the manual tests described earlier. The dental putty was attached to the incisors and the canine teeth as the test films were inserted. After the dental putty hardened, the test films were removed, and more putty was inserted between the molars. After the putty hardened completely, the rough margins were removed.

The patients were asked to visit the clinic again for the manufacturing of a new orthopedic apparatus when the patient felt discomfort in wearing the appliance or when the symptoms were aggravated because orthopedic treatment could cause a counter-deviation on the opposite side. When it was not possible to visit the clinic due to distance or time, the patient was asked to wear a TMJ balancing appliance (TBA), in which right-left deviation was not applied.

2.1.2. Acupuncture and herbal medicine treatment

Acupuncture and herbal medicine originating from Korean medicine were applied as supplemental treatments to FCST to support the patient's stamina and reduce the symptoms of the patients. The prescription of acupuncture points were mainly consisted with Hyeopcheock points and detailed points were changed at each visit in accordance with the patient's physical condition and symptoms.

Two kinds of herbal medicine (Gami-Ukgansan and Gami-Gunjatang) were given to patients. Details of the formula were changed with the condition and symptoms of patients and listed in each case (Tables 1–3). Herbal formula were provided as a water extraction packed in the retort pouch and asked to administrate 3 times a day.

2.2. Case analysis

2.2.1. IRB approvement

For the analysis of chart, we approved to review the data in the chart by institutional review board in Korea Institute of Oriental Medicine (I-1507/003-004). The use of charts were consented by the parents because the patients were under aged.

2.2.2. Visual analogue scale (VAS) analysis

At the first visit, the patients answered the VAS questionnaires provided by the clinic, and items scoring more than 5 points were selected and submitted. The patients were asked to answer the selected questionnaires at every visit to the clinic. The questionnaires answered by the patients were used to obtain the average and check for the disappearance of the symptoms reported by the patients.

2.2.3. Video analysis

At each visit, video was obtained while the patients consulted with the doctor about any changes in their symptoms or physical conditions. The patient's physical condition and the degree of severity of their tics were analyzed by referring to the treatment imaging on the first and last days of treatment and the date of the peak of improvement or worsening of the overall symptoms from the VAS analysis. The degree of severity of the tics was evaluated by recording the number of seizures per minute after classifying all of the tics into vocal tics and

Table 1 – Information of Herbal Formula Applied to the Patient

Date	Formula	Ingredients
2012.10.06–2013.12.30	Gami-Ukkansan	Pinelliae Tuber 160 g, Cnidii Rhizoma 100 g, Glycyrrhizae Radix et Rhizoma 80 g, Citri Pericarpium 120 g, Cyperus rotundus180 g, Phyllostachyos Caulis in Taeniam 60 g, Platycodonis Radix 46 g, Perillae Folium 80 g, Zizyphi Semen 400 g, Zingiberis Rhizoma Recens 60 pieces, Zizyphi Fructus 40ea

Table 2 – Information of Herbal Formula Applied to the Patient

Date	Formula	Ingredients
2013.12.14–2014.06.19	Gami-Ukkansan	Pinelliae Tuber 160 g, Cnidii Rhizoma 100 g, Glycyrrhizae Radix et Rhizoma 80 g, Citri Pericarpium 120 g, Cyperus rotundus 180 g, Phyllostachyos Caulis in Taeniam 60 g, Platycodonis Radix 46 g, Perillae Folium 80 g, Zizyphi Semen 400 g, Zingiberis Rhizoma Recens 60 pieces, Zizyphi Fructus 40ea
2014.06.20–2014.07.10	Gami-Ukkansan (dose changed)	Pinelliae Tuber 240 g, Cnidii Rhizoma 150 g, Glycyrrhizae Radix et Rhizoma 120 g, Citri Pericarpium 180 g, Cyperus rotundus 270 g, Phyllostachyos Caulis in Taeniam 90 g, Platycodonis Radix 68 g, Perillae Folium 120 g, Zizyphi Semen 600 g, Zingiberis Rhizoma Recens 80 pieces, Zizyphi Fructus 60 ea

Table 3 – Information of Herbal Formula Applied to the Patient

Date	Formula	Ingredients
2013.03.03–2013.06.14	Gami-Ukkansan	Pinelliae Tuber 160 g, Cnidii Rhizoma 100 g, Glycyrrhizae Radix et Rhizoma 80 g, Citri Pericarpium 120 g, Cyperus rotundus 180 g, Phyllostachyos Caulis in Taeniam 60 g, Platycodonis Radix 46 g, Perillae Folium 80 g, Zizyphi Semen 400 g, Zingiberis Rhizoma Recens 60 pieces, Zizyphi Fructus 40ea
2013.06.15–12.13	Gami-Gunjatang	Pinelliae Tuber 180 g, Poria 120 g, Ginseng Radix 80 g, Glycyrrhizae Radix et Rhizoma 80 g, Zingiberis Rhizoma 160 g, Agastachis Herba 120 g, Amomi Fructus Rotundus 120 g, Zingiberis Rhizoma Recens 60 pieces, Zizyphi Fructus 40 ea

motor tics, based on the entire imaging data. At that time, continuous movements were recorded as one seizure, and a seizure at different parts with a time lag within that seizure was recorded as a separate seizure.

2.2.4. The Yale Global Tic Severity Scale (YGTSS)

The Korean translated version of YGTSS[16] was used to measure the severity of tic symptoms. The parents were asked to observe their children and check the lists on the YGTSS every 7 days (case 2) or 90 days (cases 1 and 3), and they were asked to submit the YGTSS checklist to the clinic. Collected checklists were coded as numbers and analyzed.

3. Cases

3.1. *Case 1*

The age of the patient was 15 years old when she visited the clinic. Her parents reported that tics had started at the age of 6 years old and that she had been treated with various types of Western medicine for 5 years and traditional medicine for the past 6 months. They also reported that prior treatment made resulted in slight improvements in symptoms. There was no medical or family history reported.

At the first visit, the patient had difficulty eating food due to continuous tic seizures and was severely underweighted because of the disturbance from the tic seizure. The patient had continuous tics with durations of 2–3 seconds, which resembled seizures in the upper arm, shoulder, neck area, and upper body, although the symptoms had been slightly improved by previous treatments at a Korean medicine clinic. The range of movements during the tic seizures was not great, and small convulsions occurred continuously. In addition to the symptoms directly related to the tics, the patient complained about a sensitive personality, musculoskeletal inconvenience, dizziness, memory problems, chronic fatigue, dazzling and frequent inflammation inside the throat.

After manual testing for applicability, the indication for FCST was confirmed, a customized oral correction device was applied, and herbal medicine and acupuncture treatment were administered. The imaging analysis undertaken prior to treatment showed 9.7 tic seizures per minute, without vocal tics.

The herbal formula Gami-Ukkansan were applied to the patient and its composition is described in Table 1.

To briefly explain the change of symptoms prior to the detailed explanation (Fig. 1), the symptoms began to decrease immediately and decreased until 42 days of treatment and fluctuation of symptoms were consisted until about 300 treatment day. The treatment ended in 405 days with the loss of most symptoms.

The most impressive part of the change was the condition of patient. At the first visit of patient, she was very underweighted and depressed because of the tic seizure disturbed most of the patient's behaviors. The patient recovered physically and psychologically through the treatment sessions and she recovered enough to make fun with the clinician.

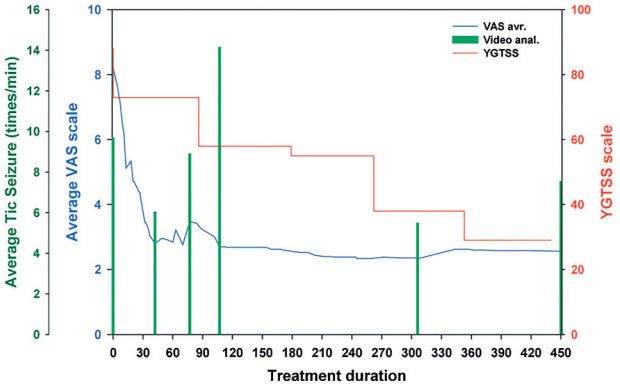

Fig. 1 – The changes of symptoms in case 1. In this figure, we briefly described the change of symptoms; a blue line shows the VAS scale; red line shows the YGTSS scale; the green bars shows the result of video analysis. We can see the strength of symptoms were decreased continuously regardless of the phase.

The video analysis showed the intensity of seizure was decreased spontaneously regardless of the frequency of seizure. The YGTSS score decreased continuously throughout the treatment.

3.2. Case 2

The age of the patient was 12 years old, and tic disorder started at 8 years old. He had been treated with acupuncture for 5 years and did not experience significant improvement. He also underwent surgical treatment for torticollis 2 years before the visit to the clinic and reported that most of the symptoms of torticollis had disappeared. There was no medical history or family history reported.

At the visit, the patient was suffering from Tourette syndrome in a combined form of vocal tics and motor tics. With the vocal tics, the patient's mother reported that the patient made an 'Ah!' sound or swore; however, video analysis showed that the patient mainly made a clicking or tick sound using the tongue. With the motor tics, movement of the neck, face, and upper arms was observed, and according to the report submitted by the parents, the patient sometimes moved his legs. In addition to the symptoms directly related to the tic, the patient complained about frequent stomatitis.

Before treatment, the frequency of tic seizures was 3.3 vocal tics per minute and 16.1 motor tics per minute, that is, a total of 19.4 tic seizures per minute. After a manual test, the FCST indication was confirmed; thus, a customized oral correction device was applied, and herbal medicine and acupuncture treatment were applied.

The herbal formula Gami-Ukkansan were applied to the patient and its composition is described in Table 2. Herbal formula were not applied to the patient after the application of high dose (1.5 times higher) medication between 2014.06.20 and 07.10.

To summarize the change of symptoms prior to the detailed explanation, this case showed the most fluctuation of the three cases in this article. The symptoms began to decrease immediately and decreased until 33 days of treatment and fluctuation of symptoms were consisted until about 235 treatment day. The treatment ended in 357 days with the loss of most symptoms.

This case showed most fluctuation of symptoms throughout the treatment. VAS score showed rapid decrease of self-recognizing symptoms; however, he showed strongest fluctuation of symptoms compared to other cases. We considered this strong fluctuation of VAS score is caused by personal character (only this patient was young boy), not the difference of the superiority or treatment methods (Fig. 2).

3.3. Case 3

The patient was 9 years old when she visited the clinic. Tic disorder started at the age of 7 years old, and no treatment had been applied because the parents believed that it was not severe and would disappear soon. She reported that there was no related family history. She experienced a car accident 4 years earlier, and she had experienced no severe injuries.

At the first visit, tic seizure had the form of movement of the neck, head, and shoulders at the same time, and the rate

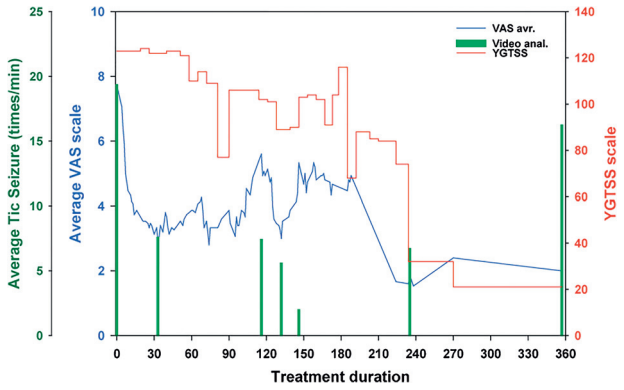

Fig. 2 – The changes of symptoms in case 2. In this figure, we briefly described the change of symptoms; a blue line shows the VAS scale; red line shows the YGTSS scale; the green bars shows the result of video analysis. We can see the tendency of decreasing strength of symptoms regardless of the phase. In case 2, the changes of the symptoms showed more unstable form compared to other cases.

of tic seizures was 1.8 per minute. According to the report by the parents, the patient moved because she felt abnormal sensations in the arms, neck, and shoulders. The patient complained about dizziness and frequent ear, nose and throat symptoms, including sinusitis, allergic rhinitis and tonsillitis, in addition to the symptoms directly related to the tic disorder.

The herbal formula Gami-Ukkansan and Gami-Gunjatang were applied to the patient and its composition is described in Table 3. The formula has changed from Gami-Ukkansan to Gami-Gunjatang, which can preserve stamina while it also partly has the stabilizing effect of Gami-Ukkansan, in the consideration of the patient's low stamina.

To summarize the change of symptoms prior to the detailed explanation, the patient complaint less severe symptoms than other two patients and showed fast and stable progress or treatment. The symptoms began to decrease immediately and decreased until about 30 days of treatment and minor fluctuation of symptoms were consisted until about 180 treatment day. The treatment ended in 457 days with the loss of most symptoms and patient kept tic with very low frequency until about 450 days.

This case, had less severe symptoms compared to other two cases. The most different part is about the fluctuation. The fluctuation observed after the rapid decrease of symptoms is barely observed in this case and the symptoms decreased spontaneously. Most of symptom disappeared in about 330 days (Fig. 3).

4. Discussion

Exposure response prevention (ERP) and habit reversal training (HRT) have been recommended as the first-line treatment for chronic tic disorder or TS because of the similar effects and less severe adverse events compared to medication treatment.[17] Another systematic review recommended only one medication type, α2-adrenergic receptor agonists (clonidine, guanfacine), at only a moderate level after comparing

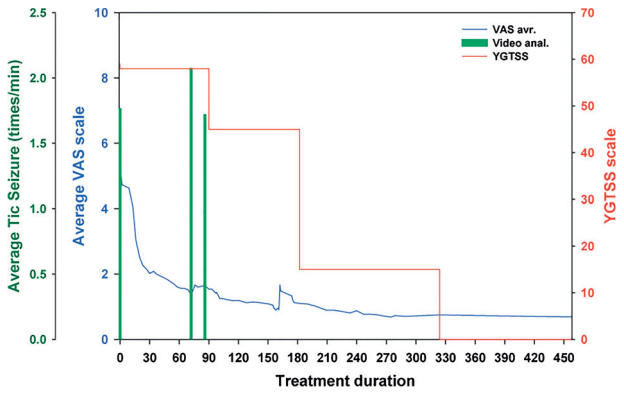

Fig. 3 – The changes of symptoms in case 3. In this figure, we briefly described the change of symptoms; a blue line shows the VAS scale; red line shows the YGTSS scale; the green bars shows the result of video analysis. We can see the strength of symptoms were decreased continuously regardless of the phase.

the balance between the benefits and adverse effects of 25 medications.[18]

FCST, an orthopedic application based on the traditional Korean medicine and applied kinesiology has been applied in these cases. In accordance with the FCST theory of treatment mechanisms, orthopedic stimulation of the TMJ can result in correction of herniated bones in the skull and cervical vertebra 1 and 2 or expansion of the TMJ inter-joint space. Those orthopedic corrections have been indicated as the reasons for efficacy in FCST theory because they can reduce the resistance of neuronal tracks.

In the cases, we performed three assessments: VAS, YGTSS and video analysis.

The VAS was performed by the patients themselves and thus was very subjective, but it could cover most of the moments. YGTSS was performed by the patient's parents to record more objective data. The results were more detailed and objective, and the duration of observation was expected to be reduced. We also considered that the YGTSS recorded by parents might have been affected by the child-parent relationship or expectations about the treatment, whether it is positive or negative. Video analysis was performed by researchers independent from the clinician, patients or parents to minimize bias from the relationships between people. In addition, the analysis was performed immediately after all treatment sessions ended to minimize the effects of time. Thus, we considered that video analysis was the most objective assessment, even though it covered only a few minutes recorded in the video clips. In this report, we considered all three of these assessments to ensure the objectivity and coverage when we analyzed the changes in the symptoms of tic disorder.

We could observe that the orthopedic treatment (FCST) decreased the symptoms of tic disorder in all three cases, in accordance with the three assessments described in the prior sentence. We could divide the treatment period into three phases. In every case, VAS scale was reduced significantly in 30 days from the first day of treatment (1st phase) and the video analysis also showed significant reduction of seizure qualitatively. Between 30 and 240–300 treatment days (2nd phase), we could observe the retention of treatment or subtle improvements in symptoms, and after the 2nd phase (final phase), most of the symptoms were dramatically reduced. We concluded that there might exist two or more working mechanisms, while treating tic disorder with the combination of orthopedic intervention, acupuncture and herbal formula caused the phases to appear.

Acupuncture treatment and herbal formula were also applied for these patients. A recent systematic review of the efficacy of acupuncture treatment for tic disorder reported that the short-term efficacy of acupuncture treatment was better than Western medicine[19] but the patients in this case report were treated with a long-term strategy. Another systematic report of the efficacy of herbal medicine reported that herbal medicine had only potential efficacy for tic disorder.[8] We concluded that acupuncture treatment and herbal formula might have worked as an adjuvant treatment to orthopedic treatment by reducing the symptoms temporally or improving the patients' physical conditions.

We searched about the prior study which treated tic/Tourette syndrome with orthopedic methods; however, we could not find any article researched the relationship between orthopedic treatment and tic disorder. Thus, we could state this report has novel finding that the orthopedic treatment could be used to control tic disorder or Tourette syndrome.

In this case report, we attempted to separate the clinician and patient from the researcher to improve the objectivity, and the results from the researcher, parents and patients had similar patterns. As a result of observation, combination treatment of orthopedic application and acupuncture and herbal medicine had a significant tendency of reduce the symptoms of tic disorder in both the short and long term, despite a lack of statistical analysis, because of the strong similarity among the three cases. In the future, we recommend large-scale clinical trials to ensure the statistically significant efficacy of single treatment with orthopedic techniques for tic disorder.

Conflict of interest

The authors declare no conflict of interest.

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