

**LETTER TO THE EDITOR**

Blacksmith's Dystonia Is Another Task-Specific Dystonia: From Past to Present

Min Seung Kim,^{1,2} Don Gueu Park,¹ Jung Han Yoon¹¹Department of Neurology, Parkinson Center, Ajou University School of Medicine, Suwon, Korea²Department of Neurology, Hallym University Dongtan Sacred Heart Hospital, Hallym University College of Medicine, Hwaseong, Korea

Dear Editor,

Task-specific dystonia (TSD) is a form of primary focal dystonia triggered by selective motor activities that usually involve highly skilled and repetitive actions.¹ Writer's cramp and musician's dystonia are the most common forms of TSD, and various types of TSD associated with patient occupations have been reported. Several occupations have disappeared over time and are no longer present in the modern era. Here, we report a patient with proximal arm dystonia triggered by using a hammer associated with his unique and traditional occupation, a blacksmith.

A 60-year-old right-handed male blacksmith presented with a 3-year history of unintentional clonic extension of his right arm when flexing his elbow to swing a hammer. He had been working traditionally as a blacksmith for 30 years. He molded iron by striking heated metal with a 3.3-pound hammer for at least 3–4 hours a day, producing sickles, hoes, or kitchen knives. Early in the disease course, clonic arm spasm was triggered only when he used the hammer. However, over the ensuing year, symptoms progressively spread to other daily activities requiring elbow flexion, such as tooth brushing or writing (Supplementary Video 1, segment 2 in the online-only Data Supplement). His family and medical histories were noncontributory. A neurological examination did not reveal any abnormalities. When he tried to flex his elbow, 3–4 Hz abrupt flexion–extension movements of the elbow joint and simultaneous contractions of the proximal arm muscles were observed (Supplementary Video 1, segment 1 in the online-only Data Supplement); the null point was detected at full extension. A sensory trick ameliorated his symptoms wherein he would touch his wrist with his left hand (Supplementary Video 1, segment 3 in the online-only Data Supplement).

No other involuntary movements were observed in other body parts. Brain and cervical spine MRI scans were normal, as were blood tests including ceruloplasmin, copper, and thyroid hormone levels.

Needle electromyography revealed 4–6 Hz polyspikes with co-contraction of the triceps brachii and biceps brachii during elbow flexion (Figure 1). No abnormal activities were detected when the elbow was extended. He was diagnosed with focal TSD of the right arm and was treated with botulinum toxin A injection (100 units for the triceps brachii). Several weeks later, he had improved markedly (Supplementary Video 1, segment 4 in the online-only Data Supplement).

We report a case of unique occupational TSD. As blacksmith is an uncommon occupation in the modern era, this type of dystonia is rarely reported. We found the only mention of “smiths’ cramp” (in the era prior to establishment of the concept of dystonia) in a renowned textbook published in 1888, *A Manual of Diseases of the Nervous System*, by Gowers.² He described a case of smith's cramp, which was very similar to our case, as follows: “...a metal-chaser, who worked with a four-pound hammer, found that, after using it for about an hour, clonic spasm came on in the biceps, and interfered with his work” Our case is interesting for its rediscovery of smith's dystonia described 130 years ago, which has not been reported with concurrent patient video and electromyography findings.

Both genetic and environmental factors are thought to be important risk factors for TSD. As up to 25% of TSD patients had an affected family member, genetic factors affecting susceptibility to TSD were suspected, but specific causative mutations have not been identified.³ Environmental factors are diverse and het-

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Corresponding author: Jung Han Yoon, MD, PhD

Department of Neurology, Ajou University School of Medicine, 164 Worldcup-ro, Yeongtong-gu, Suwon 16499, Korea / Tel: +82-31-219-5175 / Fax: +82-31-219-5178 / E-mail: jhyoon@ajou.ac.kr

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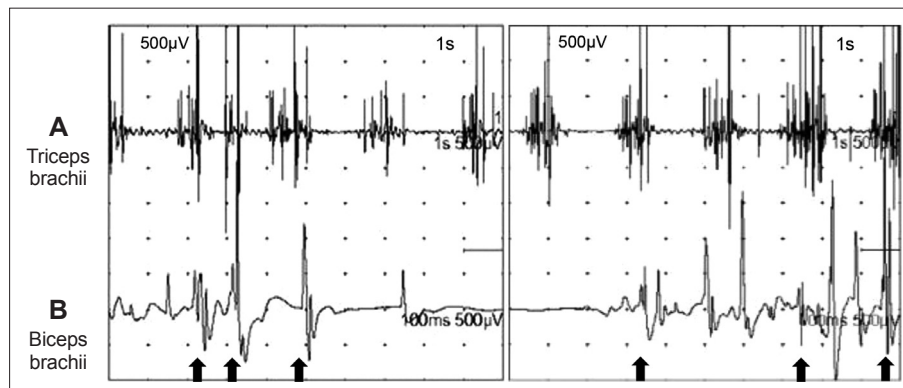


Figure 1. Needle electromyography of the right arm. When the patient tried to flex the elbow, the triceps brachii (A) and biceps brachii (B) showed simultaneous activation, suggesting co-contraction.

erogeneous between patients. Among the factors, a task demanding high spatiotemporal acuity and repetitiveness with overuse are important risk factors for TSD.⁴ This is why the majority of the occupational TSDs involved the dominant side of the hand or finger, which is usually required for repetitive, skilled motor practice. Recently, a few cases of TSD involving the proximal arm were reported.^{5,6} Likewise, iron molding with the hammer requires highly skilled action with spatial acuity and many repetitions, almost fully dependent on upper arm strength, with little intervention of wrist or hand. This case suggests that sufficient repetition and complexity of tasks possibly provokes TSD in proximal extremities. The pathophysiology of TSD remains unclear, but several mechanisms have been suggested, including reduced inhibition, motor network abnormalities, and altered neuroplasticity.³ A hypothesis of aberrant afferent inputs of basal ganglia from the cerebellum has also been suggested.⁷ Further studies demonstrating the difference in brain networks associated with the body part involved in TSD may elucidate our report. Botulinum toxin injection is the mainstay of treatment in TSD, although responses are variable between patients.⁴ In this case, despite concerns of large muscle involvement, botulinum injection worked effectively.

Ethics Statement

The authors received informed consent from the subjects.

Supplementary Video Legends

Video 1. A 60-year-old man exhibited the abrupt and large amplitude of repetitive movement triggered by elbow flexion (segment 1). These involuntary movements occurred during writing with elbow flexion (segment 2), and dystonia was attenuated when the patient contacted his left hand on his right wrist (segment 3). After botulinum toxin injection, marked improvement of dystonia was noted (segment 4).

Supplementary Materials

The online-only Data Supplement is available with this article at <https://doi.org/10.14802/jmd.22037>.

Conflicts of Interest

The authors have no financial conflicts of interest.

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Author Contributions

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ORCID iDs

Min Seung Kim	https://orcid.org/0000-0003-4397-6612
Don Gueu Park	https://orcid.org/0000-0001-8658-3523
Jung Han Yoon	https://orcid.org/0000-0001-6180-9848

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