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Myotomy and Selective Peripheral Denervation Based on ^{18}F-FDG PET/CT in Intractable Cervical Dystonia: A Case Report

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Abstract

Cervical dystonia, characterized by the involuntary contraction of cervical muscles, is the most common form of adult dystonia. In a patient with intractable cervical dystonia, we carried out a myotomy of the left obliquus capitis inferior and selective peripheral denervation (SPD) of the posterior branches of the C3-C6 spinal nerves based on preoperative 18F-fluorodeoxyglucose (^{18}F-FDG) positron emission tomography/computed tomography (PET/CT). The patient was a 65-year-old, right-handed man with an unremarkable medical history. His head rotated involuntarily to the left. Medication and botulinum toxin injections were ineffective, and surgical treatment was considered. ^{18}F-FDG PET/CT imaging revealed FDG uptake in the left obliquus capitis inferior, right sternocleidomastoideus, and left splenius capitis. Myotomy of the left obliquus capitis inferior and SPD of the posterior branches of the C3-C6 spinal nerves was performed under general anesthesia. During the 6-month follow-up, the patient's Toronto Western Spasmodic Torticollis Rating Scale score improved from 35 to 9. This case shows that preoperative ^{18}F-FDG PET/CT is effective in identifying dystonic muscles and determining the surgical strategy for cervical dystonia.

Keywords: ^{18}F-FDG PET/CT, cervical dystonia, myotomy, selective peripheral denervation

Introduction

Cervical dystonia is the most common form of adult dystonia, which is characterized by the involuntary contraction of cervical muscles. Botulinum toxin (BT) injections are the first-line treatment; however, BT injections are not always the most appropriate treatment.[1,2] To maximize the efficacy of BT injections for cervical dystonia, dystonic muscles should be identified.[3] Dystonic muscles are identified based on clinical symptoms and physical and clinical examination findings. During clinical examination, electromyography (EMG) is employed to identify the dystonic muscles; however, needle EMG is invasive and painful.[4] 18F-fluorodeoxyglucose (^{18}F-FDG) positron emission tomography/computed tomography (PET/CT) is a noninvasive modality that has been used to identify dystonic muscles in cervical dystonia.[5,6] Recently, single photon emission computed tomography (SPECT/CT) was reported to successfully identify dystonic muscles.[7] These examinations support the identification of dystonic muscles and increase the efficacy of BT injections.[3,8] However, the effect of BT injections is transient, and patients with cervical dystonia require multiple injections. For intractable cases and patients with unsatisfactory results, surgical treatments, including selective peripheral denervation (SPD) and stereotactic surgery, should be considered.[9] Preoperative clinical examinations, including ^{18}F-FDG PET/CT and SPECT/CT, can be useful in identifying the dystonic muscles to determine the target nerves and muscles. However, reports on the application of ^{18}F-FDG PET/CT and SPECT/CT before SPD are lacking.[10] This study is the first report of myotomy and SPD in a patient with intractable cervical dystonia who was assessed using preoperative ^{18}F-FDG PET/CT.

Case Report

The patient was a 65-year-old, right-handed man with

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Fig. 1 Improvement of the patient's cervical posture following selective peripheral denervation. Neutral cervical posture before the surgery (a) and after the surgery (b).

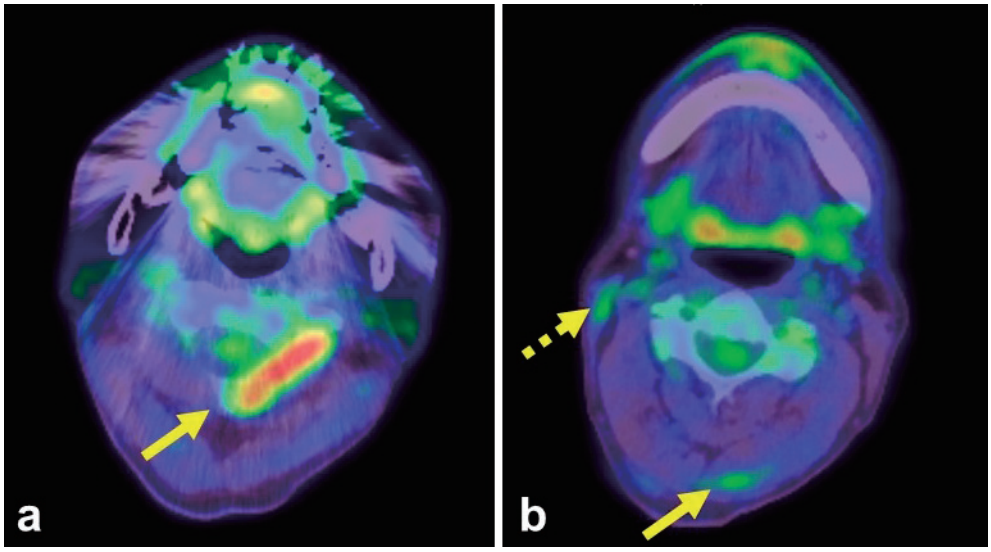

Fig. 2 18F-fluorodeoxyglucose (^{18}F-FDG) positron emission tomography/computed tomography axial imaging. The image shows abnormally high FDG uptake in the left obliquus capitis inferior (arrow) (a), right sternocleidomastoideus (dotted arrow), and left splenius capitis (arrow) (b).

an unremarkable medical history. At the age of 61, his head rotated involuntarily to the left side, and he was diagnosed with cervical dystonia. He was treated with trihexyphenidyl, clonazepam, and BT injections; however, these were ineffective. The patient was referred to our hospital, where he planned to undergo SPD. The Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) score (range 0-85) was 35 (severity scale, 16/35; disability scale, 10/30; pain scale, 9/20) preoperatively (Fig. 1). ^{18}F-FDG PET/CT was performed to identify dystonic muscles. ^{18}F-FDG (238 MBq) was injected intravenously at rest. The patient's plasma glucose level was 114 mg/dL. ^{18}F-FDG PET/CT revealed FDG uptake in the left obliquus capitis inferior, right sternocleidomastoideus, and left splenius capitis (Fig. 2). The maximum standardized uptake value for each muscle was 5.92, 2.16, and 4.70, respectively. The left obliquus capitis inferior and left splenius capitis were the main dystonic muscles. We performed a myotomy of the left obliquus capitis inferior and SPD of the posterior branches of the C3-C6 spinal nerves based on the ^{18}F-FDG PET/CT imaging.

The patient was placed in a prone position. A midline skin incision was made from the inion to the C6 spinous process. We confirmed the left obliquus capitis inferior

and performed a myotomy of the muscle. Next, the space between the semispinalis capitis and cervicis was dissected to expose the posterior branches of the C3-C6 spinal nerves. Postoperatively, the head rotation improved. The patient was discharged 7 days postoperatively. During the 6-month follow-up, his TWSTRS score improved to 9 from 35 (Fig. 1).

Discussion

BT injections are the first line treatment for medically intractable cervical dystonia; however, some cases are refractory to the injections.[9] Comella et al. reported that 25% of patients with cervical dystonia who were managed with BT injections did not improve and 13% deteriorated.[11] Surgical treatment strategies, including SPD and stereotactic surgeries are often considered because of the possibility of failure and the transient effectiveness of BT injections.[12,13] Deep brain stimulation (DBS) is a highly efficacious treatment for dystonia. However, it has several drawbacks DBS requires periodic battery replacement and the adjustments of stimulation parameters for effective results. Additionally, it carries the risk of hardware-related complications such as infections, device malfunction, and lead migration. In the present case of cervical dystonia, the symptoms were localized. SPD can avoid not only hardware-related complications but also other DBS-related disadvantages, including the risk of intracranial hemorrhage.[14] We chose to perform SPD because it is a more cost-effective alternative and does not require postoperative management. We have elected to perform SPD. The most frequently reported complication associated with SPD is sensory disturbance.[14] To minimize this, we chose the option of myotomy without denervation of the C1-C2 nerve root. SPD is an effective treatment for cervical dystonia.[13,15] The identification of dystonic muscles is essential for maximizing the efficacy of cervical dystonia treatment.[3] The dystonic muscles are identified based on clinical symptoms and physical examination; however, they can also be detected by EMG, ^{18}F-FDG PET/CT and SPECT/CT.[5-7] Jang et al. reported that the sensitivity, specificity, and accuracy of ^{18}F-FDG PET/CT for localizing dystonic muscles were 76%, 92%, and 88%, respectively.[5] In the present case, abnormal FDG uptake in the left obliquus capitis inferior was detected on preoperative ^{18}F-FDG PET/CT. The obliquus capitis inferior is a key muscle in cervical dystonia, especially, in torticaput.[16] Su J et al. reported that the obliquus capitis inferior was involved in 70% of torticaput cases based on their SPECT/CT findings.[16] ^{18}F-FDG PET/CT and SPECT/CT effectively identify the affected muscles, including the deep muscles like the obliquus capitis inferior. Without a thorough clinical examination, such as ^{18}F-FDG PET/CT or SPECT/CT, determining the involvement of the deep muscles is challenging. In our case, the main dystonic muscles were the left obliquus capitis inferior and left splenius capitis, based on the ^{18}F-FDG PET/CT study. Thus, we performed a myotomy of the left obliquus capitis inferior and SPD of the posterior branches of the C3-C6 spinal nerves that innervate the splenius capitis. The C1-C2 anterior rootlet innervates obliquus capitis inferior. Dystonic contraction of the obliquus capitis inferior is effectively treated via an intradural rhizotomy of the anterior C1 and C2 nerve roots; however, intradural rhizotomy has a risk of postoperative complications, such as cerebral spinal fluid leakage.[14] Myotomy is believed to be safer and less invasive than intradural rhizotomy.

In conclusion, we present a case of myotomy and SPD in a patient with cervical dystonia who was assessed using preoperative ^{18}F-FDG PET/CT. To identify the dystonic muscles and determine the target muscles and nerves, preoperative ^{18}F-FDG PET/CT was used. Further investigations are required to provide evidence for the effectiveness of preoperative and postoperative ^{18}F-FDG PET/CT in cervical dystonia.

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Ethical Considerations

The authors obtained the informed consent of this patient.

Conflicts of Interest Disclosure

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