Successful pallidal surgery for tardive stereotypy

A video case report

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Tardive dyskinesia is a movement disorder presenting choreoathetoid involuntary movements developed after chronic exposure to dopamine receptor blocking agents.¹ Tardive syndrome is used as a term encompassing all tardive involuntary movements,² while classical tardive dyskinesia is not included in tardive syndrome in DSM-5.¹ Tardive stereotypy, which is composed of repetitive, ritualistic, or mannerism involuntary movements,³ is one of the forms of tardive syndrome and stereotypy frequently appears as a fragment of movement in tardive dyskinesia. Previous reports have shown that internal globus pallidus (GPi) surgery is highly effective for tardive dyskinesia.^{4,5} We present a case of tardive stereotypy in which pallidotomy and pallidal deep brain stimulation (DBS) abolished the stereotypy.

PRACTICAL IMPLICATIONS

Consider pallidal surgery as a treatment option for tardive stereotypy.

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Case report

A 73-year-old woman presented with involuntary movements in the face, mouth, and shoulder, along with spontaneous stepping. She had undergone trans-thoraco-abdominal esophagectomy for treatment of esophageal cancer 8 years prior; she then underwent esophageal reconstruction 3 years ago. She took antiemetics, metoclopramide 15 mg/d, or prochlorperazine mesilate 15 mg/d for 1 year after esophagectomy. Her gait gradually became short-stepped with postural instability, and involuntary movements appeared 2 years ago. Discontinuation of antiemetics and introduction of haloperidol 1.5 mg a day did not improve the involuntary movements. Neurologic examination at the first visit to our department revealed stereotypy, which consisted of repetitive synchronized forehead contraction, mouth opening, shoulder elevation, deep inspiration, and unsuppressed alternate stepping during standing (video 1). Mild tongue choreic movement also existed. In addition, parkinsonism was recognized: symmetrical mild bradykinesia and muscular rigidity, short-stepped gait, and postural instability were present. Her mental state was normal. The results of routine blood tests were not remarkable, and her brain MRI showed mild cerebral atrophy without degenerative signs in the brainstem and basal ganglia.

As the movement symptoms strongly distressed the patient, we performed bilateral GPi surgeries after informed consent. The former esophageal surgery limited the space for implantation of 2 pulse generators in her chest wall. We then performed GPi DBS on the right side (Medtronic [Langhorne, PA] DBS lead, model 3387) and pallidotomy on the left side based on the presurgical MRI and microrecording. A postoperative MRI scan revealed an oval coagulation within the posterolateral part of the left pallidum, and the lead tip on the right side was located just on the optic tract at lateral 22 mm and 2 mm anterior to the midcommissural point (figure). The best improvement was obtained by monopolar stimulation using the most dorsal contact as cathode and the pulse generator as anode, with a stimulation frequency of 130 Hz, pulse width of 60 μ s, and >3.4 V.

Involuntary movements were abolished after pallidal surgeries (video 2). Scores on the Abnormal Involuntary Movement Scale (total scores of categories I, II, and III) decreased from 13 before surgery to 0 after surgery. When stimulation of the right GPi was turned off, involuntary

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(A) Axial and (B) frontal sections, 6 days after internal globus pallidus (GPi) surgery. A coagulation lesion with surrounding edema is located in the left GPi, and a stimulation lead is located in the right GPi with the tip just on the optic tract.

stepping during standing appeared only on the left side. The gait disturbance and postural instability improved after surgery, but transiently, and these symptoms responded little to levodopa. The effect on the involuntary movements was stable over the follow-up period, which has been 2 years.

Discussion

The patient had medication-induced tardive syndrome, mainly stereotypy, and the causal drugs were antiemetics, metoclopramide, and prochlorperazine. The common pathophysiology underlying tardive dyskinesia has been suggested to be upregulation and supersensitivity of striatal dopamine receptors. A line of evidence suggests that alterations of striatal neuron function contribute to stereotypy.⁶ The striatum comprises the patch (striosome) and matrix compartments, and stereotypy may be related to preferential activation of the patch compartment of the striatum.⁶

There have been reports on successful treatment of tardive dystonia and dyskinesia with pallidal surgery.^{4,5} The results of our patient also suggest that pallidal surgery is a promising treatment option for tardive stereotypy. Evidence suggests that the GPi is a key nucleus for the development of choreic and dyskinetic movements. An irregular, or bursting and pausing, firing pattern in the GPi has commonly been observed in hemiballismus patients,⁷ and correlation between the neuronal activity and EMGs has been observed in some GPi neurons of a patient with hemiballismus.⁷ The results of our case suggest that tardive stereotypy shares a common neural mechanism in the GPi with tardive dyskinesia, and

the causal abnormality of pallidal activities may be produced by the altered neural activities of the striatum. It is highly probable that pallidal surgery abolishes stereotypy by blocking the causal neural activities of the GPi.

Author contributions

T. Hashimoto: design and drafted and revised the manuscript for content. T. Isobe: drafted the manuscript for content. T. Goto: drafted the manuscript for content.

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