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Short communication

Deep brain stimulation of the internal globus pallidus for disabling haloperidol-induced tardive dystonia. Report of two cases

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ABSTRACT

Aim: Tardive dystonia (TD) represents a side effect of prolonged intake of neuroleptic drugs. TD can be a disabling movement disorder persisting despite available medical treatment. Deep brain stimulation (DBS) has been reported successful in this condition although the number of treated patients with TD is still limited to small clinical studies or case reports. In this study, we present 2 additional cases of patients after bilateral globus pallidus internus (GPi) stimulation.

Methods: The formal assessment included the Burke–Fahn–Dystonia Rating Scale (BFMDRS). The preoperative and postoperative functional and motor parts of this scale were compared in each patient. The postoperative assessments were done every 6 months.

Results: Both patients underwent successful bilateral GPi DBS for TD. The postoperative motor score improved by 78% at 24 months in patient 1 and 69% at 12 months in patient 2. There were no surgical or hardware-related complications over follow-up period.

Conclusion: Our experience indicates that bilateral GPi DBS can be an effective treatment for disabling TD. The response of TD to bilateral GPi DBS is very rapid and occurs within days after the procedure.

1. Introduction

Prolonged exposure to neuroleptic drugs may induce tardive movement disorders. It is estimated that in patients who take neuroleptic drugs over extended periods of time, tardive dyskinesia and tardive dystonia (TD) have an average prevalence of 15–20% and 1–4%, respectively [1]. Apart from physical impairment and pain, both tardive movement disorders cause emotional and social distress in patients with

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previous psychiatric background. Medical therapies have little effect on patients with TD. Therefore, surgical therapies for these patients are warranted.

Pallidal DBS has been demonstrated to be effective not only in medically refractory primary generalized dystonia, but also in segmental or even focal dystonia [2]. The experience of the efficacy and safety profile using pallidal DBS in TD patients has been far less researched. The underlying reason may be the fact that patients diagnosed with secondary dystonia generally fare less well after pallidal DBS than patients with primary generalized dystonia. Moreover, psychiatric comorbidity is considered to be a contraindication for DBS surgery by many investigators. These two main reasons may explain why the experience of DBS surgery in TD remains limited to small studies or case series [3–7]. Therefore, the main objective of this short communication is to present the results (12–24 months after surgery) in 2 patients with disabling haloperidol-induced TD.

2. Material and methods

Two men were included in this study who suffered from TD secondary to haloperidol exposure. The diagnosis of TD in both patients followed recommendations and diagnostic criteria provided by Adityanjee and co-authors [1]. Both patients had a history of insufficient improvement in response to medication in appropriate dosages. Family history regarding movement disorders in both patients was negative. Brain MRI scans of both patients were normal. Preoperative assessment included the disability and motor parts of the Burke–Fahn–Marsden Dystonia Rating Scale (BFMDRS). Postoperative assessment was performed at 6 and 12 months in both patients and 24 months in patient 1. The postoperative BFMDRS scores in stimulation on condition were compared to the preoperative BFMDRS scores. After expressing a written informed consent the patients were scheduled for bilateral pallidal DBS. Both patients underwent bilateral implantation of DBS leads (Model 3387, Medtronic Mineapolis, MN, USA) in the posteroventral lateral GPi under general anaesthesia in the Neurosurgical Department starting from May 2012. The IPGs (Activa SC 37603, Medtronic, Mineapolis, MN, USA) were implanted during the same operative session.

2.1. *Subject 1*

The first patient was 41-year-old right handed man diagnosed with depression at the age of 35 (Table 1). The patient's depression had been treated with neuroleptic drugs including risperidone, pimozide and subsequently haloperidol. At the age of 37, after 3 months of haloperidol intake the patient experienced slight involuntary backwards movements of his neck during cycling and after walking short distances. His neurological condition rapidly deteriorated. Within the 2 following months the dystonic movement affected predominantly craniocervical musculature, upper limbs, especially the upper limb girdles more pronounced in the left than right arm. There was evident dystonic posturing of his upper thoracic spine region. The patient was diagnosed with generalized dystonia induced by neuroleptic drug. Shortly after the diagnosis all neuroleptic agents were withdrawn. The patient was put on tetrabenazine, trihexyphenidyl, and clozapine with only slight and transient improvement in his dystonia. The patient's gait was severely compromised by retrocollis aggravated even by walking short distances. Botulinum toxin injections to posterior cervical muscles provided modest benefit. The patient was proposed bilateral pallidal DBS and scheduled for surgery and operated on in general anaesthesia as described above. Stimulation was initiated on the first postoperative day. After 2 days his neurological condition greatly improved. The patient experienced only slight retrocollis and trunk dystonia while walking distances much longer than before surgery. The dystonic movements of his arms disappeared completely. The clinical improvement has been maintained up to two years postoperatively.

2.2. *Subject 2*

This 56-year-old man with 7 years history of TD was diagnosed with bipolar disorder at the age of 43 years (Table 1). Over 6 years of his psychiatric illness he had been taking benzodia-

Table 1 – Baseline clinical characteristics of 2 patients with tardive dystonia.

Patient number	Age at surgery, y/sex	Disease duration	Indication for neuroleptic treatment	Neuroleptic inducing tardive dystonia	Neuroleptics exposition, mo	Medication preoperative suppressive used for tardive dystonia	Medication postoperative	Postoperative follow-up, mo
1	41/M	4	Depression	Haloperidol	3	Tetrabenazine Trihexyphenidyl Clozapine Botulinum toxin injections	Clozazepam occasionally	24
2	56/M	7	Depression	Haloperidol	24	Clozapine Clonazepam Trihexyphenidyl Tetrabenazine Botulinum toxin injections	Clonazepam occasionally	12