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Review article

Deep brain stimulation for intractable tardive dystonia: Literature overview

Michał Sobstyl [*], Mirosław Ząbek [1]

Neurosurgical Department of Postgraduate Medical Center, Warsaw, Poland

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ABSTRACT

Background: Tardive dystonia (TD) represents a side effect of prolonged intake of dopamine receptor blocking compounds. 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. The aim of this study was to present the systematical overview of the existing literature regarding DBS for intractable TD.

Methods and results: A literature search was carried out in PudMed. Clinical case series or case reports describing the patients with TD after DBS treatment were included in the present overview. Literature search revealed 19 articles reporting 59 individuals operated for TD. GPi was the target in 55 patients, while subthalamic nucleus (STN) was the target in the remaining 4. In most studies the motor part of Burke–Fahn–Marsden Dystonia Rating Scale (BFMDRS) was improved by more than 80% when compared to preoperative BFMDRS scores.

Conclusions: The performed literature analysis indicates that bilateral GPi DBS is an effective treatment for disabling TD. The response of TD to bilateral GPi DBS may be very rapid and occurs within days/weeks after the procedure. The efficacy of bilateral GPi DBS in TD patients is comparable to results achieved in patients with primary generalized dystonia.

1. Introduction

Chronic intake of dopamine receptor blocking compounds may have as a consequence the development of tardive movement disorders including tardive dyskinesia and tardive dystonia (TD) [1]. Both tardive movement disorders cause emotional and social distress but TD develops faster, is more painful, distressing and disabling than tardive dyskinesia [1]. TD usually does not differ from focal, segmental or generalized primary dystonia. In two-thirds of all cases TD affects cervical muscles. TD treatment consists of gradual withdrawal of provoking medications and substitution of atypical neuroleptics such as clozapine or administration of tetrabenazine, dopamine agonists, and anticholinergic drugs [1]. In some cases pharmacological treatment of TD may be challenging and ineffective. Clinical similarity between TD and primary dystonia has paved the way for its neurosurgical treatment – nowadays mainly with pallidal DBS [2].

* *Corresponding author at*: 8 Kondratowicza Street, 03-242, Warsaw, Poland. Tel.: +48 22 32 65 779; fax: +48 22 326 589.
E-mail addresses: mrsob@op.pl (M. Sobstyl), zabek@mds.pl (M. Ząbek).
[1] Tel.: +48 22 32 65 779.

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Pallidal DBS has been shown effective in medically refractory primary generalized, segmental or even focal dystonia [2,3]. The experience of the efficacy and safety profile using pallidal DBS in TD patients is far less studied. The reasons for it may be the following facts. Patients diagnosed with secondary dystonia (such as hemidystonia or dystonia associated with cerebral palsy) fare less well after pallidal DBS than patients with primary generalized dystonia [4]. Psychiatric co-morbidity is considered 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 [5–14]. Therefore, the main aim of this overview was to present a systematic literature review dealing with DBS for TD. We have performed the detailed discussion on the outcomes, stimulation settings, and surgical complications as well the safety profile of bilateral pallidal stimulation in TD patients. Moreover the possible underlying effects of DBS for TD using functional imaging studies have been discussed.

2. Clinical characteristics of tardive dystonia patients treated by deep brain stimulation

Since 2001, we have identified 9 original articles, in which a total of 38 patients diagnosed with TD underwent DBS procedures [5–13]. Moreover, since 2003 in another 10 published articles reporting a total of 198 patients with various types of primary or secondary dystonia we have found additional 21 patients harboring neuroleptic-induced or drug-induced TD [3,15–23]. To our knowledge, a total of 59 patients with TD have been described in literature. In 10 articles that present the series for various types of dystonia some data regarding patients' sex, disease duration, and clinical outcome are often presented as a cumulative outcome for all dystonia patients regardless of dystonia type, which considerably handicaps the clinical analysis of patient data. Nevertheless, among 59 patients there were 27 females and 20 males; in the remaining 12 individuals the patient's gender could not be identified. The mean TD duration from diagnosis to surgery was 5.5 years ranging 0.5–23 years. The mean age at surgery was 50.2 years ranging from 28 to 76 years.

In the 9 articles reporting the outcome only for TD patients the psychiatric indication for using neuroleptic drugs was stated. To the contrary, none of the 10 articles reporting the outcome for various dystonic conditions presented the underlying psychiatric indication. Among known psychiatric indications, neuroleptics subsequently developing TD were used to treat depression in 19 patients, schizophrenia in 5 patients, and in the remaining 11 patients various psychiatric disorders including bipolar disorder, psychosis, or anxiety disorder. Interestingly, out of a total of 59 patients, only 3 patients developed TD after prolonged antiemetic treatment in which metoclopramid was administered to treat gastritis [10,13,17]. In the remaining 56 patients TD developed as a consequence of neuroleptics. The most common identifiable neuroleptic was haloperidol found in 15 patients as a causative drug for inducing TD. In general, in 42 patients specific neuroleptics were identified, whereas in the remaining 14 patients no neuroleptic drug was named. The exact time of the neuroleptic exposure was provided in 23 patients and the mean time from neuroleptic exposure to TD development was 43.9 months ranging from 3 to 300 months.

Most individuals (53 patients) underwent bilateral GPi DBS [5–13,15–23]. Moreover, in 2 additional patients unilateral GPi DBS was performed to treat TD 13. In only 4 patients bilateral STN DBS was undertaken [17,18]. In 1 patient two targets GPi and ventral intermediate thalamic nucleus (Vim) were approached [5]. The mean postoperative follow-up period was 29 months ranging from 3 to 80 months.

3. Clinical outcome of pallidal deep brain stimulation for tardive dystonia

DBS for TD was first reported by Trottenberg et al. and revealed a high efficacy of bilateral GPi stimulation in a 70-year-old patient with TD decreasing the total BFMRDS scores by 73%, whereas thalamic Vim stimulation had no effect on dystonic symptoms [5]. The same study group presented additional results in 5 consecutive TD patients decreasing the BFMDRS motor and disability scores by 83% and 94% respectively, 6 months postoperatively [6]. Gruber et al. presented the largest series to date, featuring 9 patients including 3 patients reported earlier by Trottenberg et al. [10]. In this study at the mean postoperative follow-up time of 40.7 $\pm$ 20.9 months the BFMDRS motor and disability scores improved by 83% and 68% respectively [10]. Franzini at al. [7] as well Cohen et al. [8] presented 2 patients in each study, also reporting a very favorable outcome. Sako et al. [9] operated on 6 patients with TD decreasing the BFMDRS motor and disability scores by 86 $\pm$ 14% and 80 $\pm$ 12% respectively at mean follow-up of 21 $\pm$ 18 months. Capelle et al. [11] included 2 patients with craniocervical TD and 2 patients with generalized TD. At the last follow-up of 27.3 months the BFMDRS motor and disability scores improved by 77% and 84% respectively. In the last study reported by Chang et al. [12] the BFMDRS motor and disability scores improved by 71% and 48% respectively at the last follow-up ranging from 24 to 96 months. In the recent published study by Shaikh et al. 8 patients with TD were included. The total motor BFMDRS scores were improved by 85.1 $\pm$ 13.5% at the last follow-up ranging from 12 to 60 months. The outcomes of all 9 articles are summarized in Table 1 in a chronological order, by objective results based on BFMDRS scores after bilateral pallidal DBS. Some authors used additionally Abnormal Involuntary Movement Scale (AIMS) or Extrapyramidal Symptoms Rating Scale (ESRS) which may suggest that not all patients included in the study suffered from Type I – Pure Tardive Dystonia according to the recommendations provided by Adityanjee et al. [1]. Some of these patients may have exhibited Type II Tardive Dystonia with coexisting dyskinetic movements in the same or a different body part, but dystonia was the most prominent manifestation. In the study by Trottenberg et al. [6], Gruber et al. [10], and Chang et al. [12], the above-mentioned scales like AIMS or ESRS were used in addition to the BFMDRS scores in order to rate coexisting dyskinetic movements.

In the remaining 10 articles reporting series with various types of dystonia, in 2 articles the STN was the target to treat