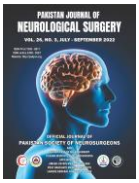

PAKISTAN JOURNAL OF NEUROLOGICAL SURGERY (QUARTERLY) – OFFICIAL JOURNAL OF PAKISTAN SOCIETY OF NEUROSURGEONS

Original Article

# Analysis of Bilateral Stereotactic Pallidotomy in Patients Presenting with Generalized Dystonia

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## ABSTRACT

**Objective:** Dystonia is an extremely painful disease-causing disability. To make the right treatment of choice dystonia should be categorized in the past, Pallidotomies were regularly performed which lead to a marked improvement in dystonia. So the rationale of the study was to find the improvement with stereotactic pallidotomy. The main aim of the study was to assess the outcome based on mean change in Burke-Marsden-Fahn Dystonia Scale (BMFDS) score after stereotactic pallidotomy in patients presenting with generalized dystonia.

**Material & Methods:** Quasi-experimental study was conducted over five years at Unit II, Department of Neurosurgery, Lahore General Hospital, Lahore. A total of 16 patients 9 were males and 6 were female meeting inclusion criteria of age 20-70years of either gender presenting with generalized dystonia for at least 6 months. Patients underwent stereotactic pallidotomy. Furthermore, Patients with coagulopathies (PT, apt > 4 sec deranged), Patients with a history of trauma, Patients with CVA (on history), and patients having intracranial pathology (trauma/hematoma) were excluded. BMFDS score was noted at baseline and after 3 months of surgery, and change in BMFDS was noted. The questionnaire was used to collect information. SPSS Version 21 was used to examine the data that had been gathered.

**Results:** The patients' average age was 47.35 and 14.40 years. There were 19 (31.67%) males and 41 (68.33%) females. The mean duration of dystonia was 15.43 ± 6.13 months. At baseline, the mean BMFDS was 49.67 ± 5.69 which was reduced to 18.03 ± 3.35. The mean change in BMFDS was 31.63 ± 6.38. There was a significant change in BMFDS ($p < 0.05$).

**Conclusion:** Hence, in patients with generalized dystonia, stereotactic pallidotomy is beneficial in lowering the BMFDS score by more than 50%.

**Keywords:** Stereotactic pallidotomy, Burke-Marsden-Fahn Dystonia Scale score, generalized dystonia.

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Date of Submission: 02-01-2023

Date of Revision: 02-02-2023
Date of Acceptance: 15-03-2023
Date of Online Publishing: 31-03-2023
Date of Print: 31-03-2023

**DOI:** 10.36552/pjns.v27i1.843

## INTRODUCTION

The movement disease known as dystonia is characterized by persistent or intermittent muscle contractions that result in aberrant, frequently repeated motions, postures, or both. It is a disorder leading to impairment. Stereotactic functional neurosurgery is a well-established procedure that is used in the treatment strategy if the medicine is ineffective. For dystonia, DBS is currently favored over lesioning methods. Thalamotomy and pallidotomy, however, served as the cornerstones of stereotactic neurosurgery treatment for dystonia from the 1950s until the 1980s.[1] For deep-seated lesions that are resistant to open surgical biopsy or resection, frame-based stereotactic biopsies have proven to be the best option for providing a histopathological diagnosis to direct subsequent treatment. Over time, both frame-based and frameless techniques have been applied to stereotactic robots.[2] In the 1990s when DBS was introduced RF lesioning almost vanished. In the modern era, most neurosurgeons working in functional neurosurgery most of them are trained in the deep brain stimulation (DBS) era and a significant amount of neurosurgeons of them have not seen lesioning procedures.[3] Levodopa-responsive dystonia is an exception to the general rule that pharmacological treatment for segmental/generalized dystonia with anticholinergic, dopamine-depleting medications (such as tetrabenazine and its derivatives), baclofen, and benzodiazepines is restricted or limited due to systemic side effects and ineffectiveness.[4] According to a recent study, individuals who first presented with dystonia symptoms did not get worse after having their devices taken out and their wounds healed. However, the research identified one patient who experienced dysarthria, swallowing issues, and a little bleeding in the lesion. Dystonic symptoms returned in two cases.[5] Further experience of the modern post-ventral pallidotomy globally in post-levodopa parkinsonian patients confirmed that it was most efficient on the 'on–off' fluctuations and the levodopa-induced dystonia and dyskinesia.[6] Bilateral STN DBS significantly reduces cervical dystonia while preventing mental decline, and a study found that STN DBS may be an effective alternative to GPi DBS for treating primary cervical dystonia[7].

The purpose of this study is to measure the mean decrease in Burke-Marsden-Fahn Dystonia Scale (BMFDS) score after stereotactic pallidotomy in patients presenting with generalized dystonia. In literature, it has been shown that pallidotomy can help reduce the severity of the disease and may relieve the patient from severe conditions by reducing the BMFDS score. But we cannot rely on the results of previous studies as those were conducted on very small sample sizes i.e., 5 & 16 cases. A study on the effects of STN deep brain stimulation in the treatment of isolated dystonia found that it was long-term tolerable and sustainedly effective.[8] Moreover, no local evidence has been found in the literature which could show that pallidotomy is an effective technique for the management of generalized dystonia. So, through this study, we want to confirm whether this procedure applies to the local population by getting the evidence by using an increased sample size as compared to previously quoted studies.

## MATERIALS AND METHOD

### Study Design and Setting

The Lahore General Hospital's Unit II, Department of Neurosurgery, was the site of this quasi-experimental Study. The scoop of 16 patients met inclusion criteria through Non-Probability, Consecutive sampling, and 95% confidence level, and taking expected change in BMFDS i.e. 19 ± 3 after stereotactic pallidotomy for generalized dystonia.

### Inclusion Criteria

Individuals included of either gender between the

ages of 20 and 70 who report generalized dystonia for at least six months.

## Exclusion Criteria

Patients with coagulopathies (PT, apt>4sec deranged), a history of trauma, CVA (on history), and intracranial pathology (trauma/hematoma) were not included in the study.

## Data Collection

From the wards of the neurosurgery department at Lahore General Hospital, 16 patients who met the enrollment criteria were accepted. That was done with informed consent. Demographic variables (name, age, gender, and site) were also noted. At baseline, the BMFDS score was noted. After taking informed consent from the patients, the patients were subjected to surgery. Regular follow-up was advised. After 3 months, patients were evaluated for BMFDS, and change in BMFDS was noted (as per operational definition). A predesigned form was used to collect information.

## Data Analysis

To analyze the collected data, SPSS Version 25 was employed. Age and BMFDS at baseline and post-operative were two quantitative variables that were provided as mean and SD. Gender and the site involved were two qualitative factors that were reported as frequency and percentage. Change in BMFDS was calculated (as per operational definition). The difference in BMFDS between baseline and after surgery was compared using a paired sample t-test. Data was stratified for age, gender, BMFDS at baseline, and duration of dystonia. Paired sample t-test was used post-stratification to compare the mean change in stratified groups. P-values under 0.05 were deemed significant.

# RESULTS

## Descriptive Statistics of the Age of Patients

Table 1 depicts that a total of 16 patients participated in the study with a maximum age of 69 whereas the least population targeted was 20. The mean age calculated was 47.35 ± 14.40.

## Comparison of BMFDS on Follow-up

Table 2 illustrates the targeted population of 16 individuals. At baseline, the mean BMFDS was 49.67 ± 5.69 which was reduced to 18.03 ± 3.35. The mean change in BMFDS was 31.63 ± 6.38. There was a significant change in BMFDS ($p < 0.05$).

## Comparison of BMFDS on Follow-up Stratified for the Duration of Dystonia

Data was stratified for the duration of dystonia. In patients having dystonia for ≤ 12 months, the mean BMFDS was reduced from 48.65 ± 4.30 to 17.70 ± 3.34. The mean change in BMFDS was 30.96 ± 4.71. There was a significant change in BMFDS ($p < 0.05$). In patients having dystonia

**Table 1:** Age Distribution.

| | | |
|---|---|---|
| Age (years) | n | 16 |
| | Mean | 47.35 |
| | SD | 14.40 |
| | Minimum | 20 |
| | Maximum | 69 |

**Table 2:** Comparison of BMFDS on follow-up.

| | Baseline | After 3 Months | Change |
|---|---|---|---|
| n | 16 | 16 | 16 |
| Mean | 49.67 | 18.03 | 31.63 |
| SD | 5.69 | 3.35 | 6.38 |
| Minimum | 40 | 12 | 17 |
| Maximum | 60 | 25 | 46 |

Paired Samples Test = 38.419
P -value = 0.000 (Significant)

> 12 months, the mean BMFDS was reduced from 50.30 ± 6.38 to 18.24 ± 3.39. The mean change in BMFDS was 32.05 ± 7.25. There was a significant change in BMFDS ($p < 0.05$).

**Table 3:** Analysis of BMFDS on follow-up stratified by dystonia duration.

| Duration (Months) | BMFDS | Baseline | After 3 Months | Change | p-value |
|---|---|---|---|---|---|
| ≤ 12 | Mean | 48.65 | 17.70 | 30.96 | 0.000 |
| | SD | 4.30 | 3.34 | 4.71 | |
| > 12 | Mean | 50.30 | 18.24 | 32.05 | 0.000 |
| | SD | 6.38 | 3.39 | 7.25 | |

## DISCUSSION

The current study compared BMFDS on follow-up stratified for the duration of dystonia. However, stereotactic pallidotomy is effective in reducing > 50% reduction in BMFDS score in patients with generalized dystonia. The mean age of patients was 47.35 ± 14.40 years. Data was stratified for the age of patients. In patients aged 20 – 45 years, the mean BMFDS was reduced from 50.38 ± 5.29 to 18.25 ± 3.15. The mean change in BMFDS was 32.13 ± 6.09. There was a significant change in BMFDS ($p < 0.05$). In patients aged 46-70 years, the mean BMFDS was reduced from 49.19 ± 5.97 to 17.89 ± 3.51. The mean change in BMFDS was 31.31 ± 6.63. There was a significant change in BMFDS ($p < 0.05$).

Contrary to our study, the ventral lateral thalamic nuclei were the site where stereotactic therapy was frequently used to treat dystonia. The most effective treatment for either primary or secondary generalized dystonia, according to Cooper, is thalamotomy. There was a significant decrease of 70% in symptoms of dystonia. Another author, Andrew et al., found no success with Cooper's method and reported only a 25% decrease in primary or secondary generalized dystonia symptoms after thalamotomy. There was a relatively high risk of postoperative dysarthria and dysphagia following bilateral thalamotomy.[9] In a different survey, no statistically significant difference in BAD scores between the DBS and pallidotomy groups could be identified (p for interaction term > 0.05).

SD resolution time following surgery (DBS 34.8 19 days, pallidotomy 21.8 20.2 days, ($p > 0.05$)) and risk difference between the two procedures (DBS vs pallidotomy 0.03; 95% CI 0.36 to 0.30) were probably not statistically different between the two procedures.[10]

Three effective dystonia treatments were identified during a recent investigation. It is expected that 90% of patients with the DYT-1 mutation, 80% of those with idiopathic GD, and those who developed later dystonia had unsatisfactory outcomes after receiving therapy with GPi DBS at a three-year follow-up. According to our research, DBS has a positive impact on patients with generalized dystonia's (GD) quality of life, and the treatment is well tolerated.[11]

In contrast to this study, another study found that patients with dystonia who underwent DBS of the GPi or VIM experienced an increase in body weight, as measured by a significant increase ratio of the BMI of about 11.5% over 6 years, with a pronounced increase during the first few months following surgery.[12] According to a study by Shiro Horisawa, bilateral stereotactic ventrolateral thalamotomy significantly reduced musicians' dystonia in both hands without any serious side effects, and the improvements persisted for a year after surgery in two participants.[13]

## CONCLUSION

In our study, stereotactic pallidotomy is effective in reducing >50% reduction in BMFDS scores in patients with generalized dystonia. Thus, it has been proved that pallidotomy can help reduce the severity of the disease and may relieve the patient from severe conditions by reducing the

BMFDS score. Now we have also got local evidence and we will implement the procedure in the local setting.

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## Additional Information


**Disclosures:** Authors report no conflict of interest.

**Ethical Review Board Approval:** The study conformed to the ethical review board requirements.

**Human Subjects:** Consent was obtained from all the patients/participants in this study.

**Conflicts of Interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following:

**Financial Relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work.

**Other Relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

## AUTHOR'S CONTRIBUTIONS

| S. No. | Author's full name | Intellectual contribution to paper in terms of |
|---|---|---|
| 1. | Muhammad Hassan Raza | Study Design, Methodology, and Data Calculation. |
| 2. | Hassan Ali Khosa &<br>Muhammad Hassan Raza | Data Analysis, Interpretation of Results, and Paper Writing. |
| 3. | Hassaan Zahid, & Adeeb-ul-Hassan | Literature Review |
| 4. | Asad Shah | Statistical Analysis. |
| 5. | Khalid Mahmood | Quality Insurer. |