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Surgical Outcome of Cervical Dystonia: A Single Center Study

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Abstract

Introduction: Dystonia is a movement disorder characterized by involuntary, sustained, and patterned muscle contractions, which cause twisting and repetitive movements. Dystonia includes various types of movement disorders such as cervical, generalized and task specific focal hand dystonia. Several potential stereotactic targets have been suggested for the treatment of dystonia such as GPi(Globus pallidus internus) and PTT (Pallidothalamic tract) for generalized and cervical dystonia and Thalamotomy for task specific focal hand dystonia.

Materials and Methods : All the patients of cervical dystonia who underwent surgical interventions in Annapurna Neurological Institute and Allied Sciences since January 2015 till December 2023 were included in our study. We performed unilateral pallidotomy or pallidothalamic tractotomy for cervical dystonia. We used ZD Fishers frame and the thermal lesioning machine of Cosman Radiofrequency (RF) generator with the lesioning electrode of 0.75 mm internal diameter and 2 mm exposed tip was used.

Results: There were altogether 42 cases of cervical dystonia. Twenty nine cases had unilateral Pallidotmy and thirteen cases had unilateral pallidothalamic tract (PTT) lesioning. The mean age was 41.49 years±12 years. The male: female ratio was 3:1. The mean percentage change in BFMDRS was 70 percent (p value<0.05) in pallidotomy group and 75% (p value<0.05) in PTT lesioning group in postoperative period. Similarly there was 60 % improvement in TWSTRSs score in pallidotomy group(p value<0.05) and 70% improvement in PTT group(p value<0.05). One case had hemiparesis, three cases had transient dysarthria and one case had hypophonia.

Conclusion: Lesioning surgery is rewarding for cervical dystonia. It is less time consuming and cheaper and it should not be abandoned. Both unilateral PTT lesioning or unilateral pallidotomy is equally effective in cervical dystonia. Though some recurrent cases may benefit from Deep brain stimulation (DBS) surgery we believe lesioning surgery is effective with good outcome.

Keywords: Cervical dystonia, Lesioning, Movement disorder

Introduction

Cervical dystonia is related to involuntary neck movements like horizontal rotation, ante- and retroflection, and lateral bending that needs a complex combination of bilateral neck muscles.[1] Surgical treatments are considered to treat cases of refractory cervical dystonia after the failure of conservative treatments, such as medications and botulinum toxin injections. Surgical treatments for cervical dystonia include selective peripheral denervation, deep-brain stimulation (DBS), and radiofrequency (RF) thermocoagulation.[2,3,4] Several potential stereotactic targets have been suggested for the treatment of dystonia such as Globus Pallidus Internus (GPi) and Pallidothalamic tract(PTT).

DBS is also a good option, but its high initial cost, hardware-related complications and regular outpatient visits must be considered when implementing it. Stereotactic ablative surgery can be as effective as DBS at less cost. In most developed countries where DBS is widely accessible, GPi ablation (pallidotomy), an alternative surgical treatment for dystonia, is rarely used. However, MR guided focused ultrasound therapy has resurged the Pallidotomy and PTT lesioning for Dystonia as well. According to limited cohort studies, pallidotomy and GPi-DBS are equally effective in treating cervical dystonia.

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Materials and Methods

Standard protocol approval, registration, and patient consent

This study was approved by the medical ethics committee of Annapurna Neurological Institute and Allied Sciences. The medical ethics committee approved a waiver of consent for the collection of data as part of the routine clinical care and quality control.

Patient data

Inclusion criteria: All the patients of cervical dystonia who underwent surgical interventions in Annapurna Neurological Institute and Allied Sciences since 2015 were included in our study.

Exclusion criteria:All the surgical cases of task specific focal dystonia and generalized dystonia have been excluded.

We did unilateral pallidotomy or pallidothalamic tract lesioning for Cervical dystonia .

Evaluation:

A systematic analysis of patient demographics, disease duration were executed.

Burke Fahn Mardsen Dystonia Rating Scale (BFMDRS) score was adopted to assess clinical outcomes both preoperatively and postoperatively. Similarly Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) was also used to evaluate the patients.

Surgical procedures

As a part of the protocol, brain MRI (3 T, Siemens) with no spacing and 3-Dimensional Volume reconstruction image was performed and it was obtained in digital imaging communication in medicine (DICOM) standard. The image was loaded in Inomed Planning Software (IPS), where the targeting and trajectory for stereotactic GPi pallidotomy was done (Fig. 1A). The anterior commissure (AC) and posterior commissure (PC) were visualized and AC- PC distance was calculated. The primary target for GPi was 18 to19.5 mm lateral, 3 mm below (caudal) and 2 mm anterior to the mid-commissural point and the secondary target was posterolateral to the primary target. The target was reconfirmed through the inbuilt Schaltenbrand atlas (Fig. 1B). We calculated PTT lesioning after visualizing the mammilothalamic tract and from the midpoint of AC-PC line.(Figure(2A). We also verified in the Schaltrenbrand Atlas(Figure2B)

Under local anesthesia, stereotactic frame (MRI compatible Zamorano-Dujovny Fisher) frame was applied and computed tomography (CT) scan of head was done (64 slice Siemens with 2 mm slice thickness) with no tilt. These images were retrieved in a DICOM format. Then the MRI and CT images were fused in the workstation and the working coordinates for the surgical procedure was extracted.

The patient was then taken to the operating room and the frame was fixed in the Mayfield. The coordinates were set on phantom to reconfirm the accuracy of target. Once confirmed, a burr hole just anterior to coronal suture was made.

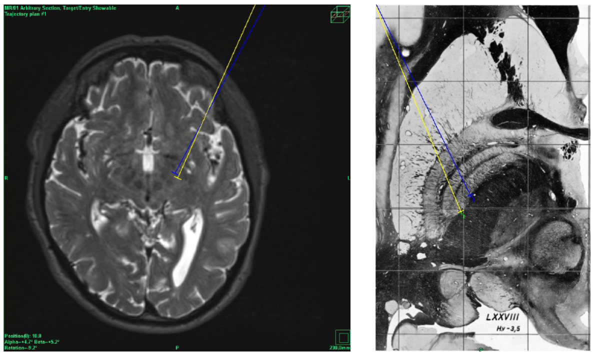

Figure 1 A&B : Targeting of left GPi and it's reverification in Schaltrenbrand Atlas

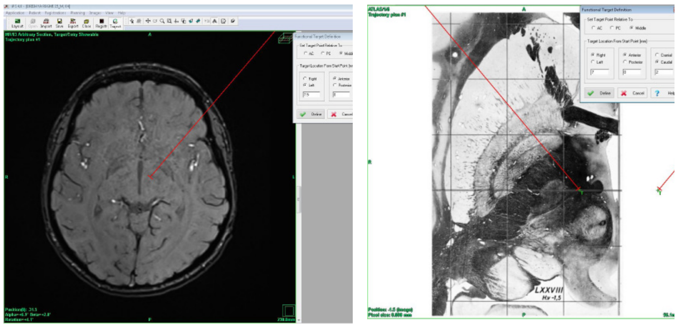

Figure 2 A&B : Targeting of left PTT and it's reverification in Schaltrenbrand Atlas

Results

Out of 88 cases of dystonia(16 generalized, 30 Task specific and 42 cervical dystonia), we included 42 cases of cervical dystonia. Out of 42 cases of cervical dystonia, 29 cases had unilateral Pallidotomy and 13 cases had PTT lesioning as shown in fig 3a&b. PTT lesioning was started since September 2023.

Patient demographics and outcome are detailed in Table 1. The mean age was 41.8 ±13 years in Pallidotomy group 40.8±8 years in PTT lesioning group. The male: female ratio was 3:1. The mean percentage change in BFMDRS was 70 percent (p value<0.05) in postoperative period in pallidotomy patients and 75% in PTT lesioning patients. Similarly, there was 60 % improvement in TWSTRSs score in pallidotomy group(p value<0.05) and 70% improvement in PTT group(p value<0.05). There was relapse of symptoms 2 cases of cervical dystonia(post pallidotomy). One case had hemiparesis, three cases had transient dysarthria. There was hypophonia in one case of cervical dystonia post PTT lesioning. There was no mortality.

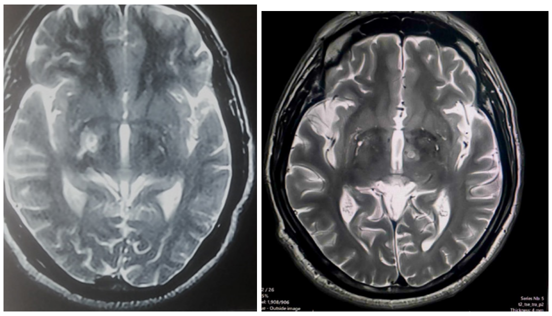

Figure 3 A&B : T2 axial MRI showing right GPI pallidotomy and left PTT lesioning

Table 1 : Demographics and surgical outcome

Target	Gpi lesioning(Pallidotomy	PTT lesioning
Number	**29**	**13**
Male: Female	3:01	3:01
Mean age	41.8±13 Years	40.8±8 Years
Mean Change in BFMDRS(Neck scale)	70%(p value <0.05)	75%(p value <0.05)
Mean change in TWSTRS	65%(p value<0.05)	70%(p value<0.05)
Mean follow up	10±2 Years	2±1 Years
Relapse	7%	None
Complication	Hemiparesis 1	Hypophonia:1
	Transient Dysarthria: 2	

Discussion

Before the advent of DBS era, various stereotactic lesioning targets were used to treat cervical dystonia, such as the zona incerta (ZI), globus pallidus internus (GPi), Forel's field H1, ventro-oral internus (VOI), and the interstitial nucleus of Cajal (INC).[5,6] These procedures were typically performed unilaterally to minimize the risk of severe complications linked to bilateral lesions.

Pallidotomy or pallidal DBS targets the posteroventrolateral part of GPi which is strongly associated with the origins of pallidothalamic fibers.[1]

Pallidothalamic tract includes the pallidothalamic fibres a major motor pathway in the brain that connects the globus pallidus internus (GPi) to the motor nuclei of the thalamus. It is composed of two main fiber bundles, the ansa lenticularis and the lenticular fasciculus, which merge in an area called Forel's field H before projecting to the thalamus. which are the merging fibers and it is more precise.

Both pallidotomy and pallidal DBS focus on the posteroventrolateral region of the GPi, which has strong connections to the origin of the pallidothalamic fibers.[7]

In our study, the mean percentage change in BFMDRS was 70 % (p value<0.05) and 75% (p value<0.05) in postoperative period in Pallidotomy group and PTT lesioning group respectively. Similarly there was 65 % improvement in TWSTRS in Pallidotomy group and 70 % improvement in TWSTRS score in PTT group.There was relapse of symptoms 2 cases of cervical dystonia in long follow up period. However, there was no relapse in PTT group. It might be because of shorter follow up of those cases.

A recent pooled meta-analysis demonstrated that bilateral GPi-DBS led to improvements of 60.4%, 54.8%, 67.7%, and 55.9% in TWSTRS total, severity, disability, and pain scores, respectively.[8] Horisawa et al showed that unilateral pallidotomy produced significant improvement in cervical dystonia, with a 47.9% reduction in the TWSTRS total score six months after surgery.[9,14] Moreover, a recent study reported that bilateral and unilateral pallidotomies reduced symptoms on the neck subscale of the BFMDRS by 73% and 50%, respectively. [10] Previous research on bilateral pallidotomy for dystonia has demonstrated remarkable outcomes, showing a 50%–90% improvement in BFMDRS scores,

which aligns with our results (70% improvement in BFMDRS).[5,11] In contrast, studies on unilateral pallidotomy for dystonia remain limited. Ondo et al. found that unilateral pallidotomy produced a 50%–73.9% improvement in BFMDRS scores among patients with trauma-induced and hereditary dystonia.[16] Similarly, Alkhani et al. documented an 84% improvement on the Unified Dystonia Rating Scale (UDRS) two years after surgery in a patient with hemidystonia of unknown cause.[13] Furthermore, While GPi-DBS is commonly used as a surgical treatment for patients with bilateral craniocervical or truncal dystonia, its use—and that of unilateral pallidotomy—in patients with midline dystonia has been rarely assessed through standardized evaluation scales.[15] Isaias et al evaluated that pallidal DBS is a safe and effective treatment for medically refractory primary generalized dystonia PGD, with improvement sustained for up to 8 years in 1 patient without significant changes in stimulation variables.[4] Hung et al reported that ten patients with severe cervical dystonia who underwent bilateral globus pallidus internus (GPi) DBS, TWSTRS severity score improved by 54.8%, the TWSTRS disability score improved by 59.1%, and the TWSTRS pain score improved by 50.4% at last follow up. [16] Martinez et al demonstrated that 80 consecutive patients with dystonia who underwent neurosurgical management by means of intrathecal pump implantation, pallidotomy or deep brain stimulation (GPi or VIM) had mean improvement in BFMDRS score among patients with primary and secondary dystonia,87.54% and 42.21%, respectively.[17] Similarly, hemidystonic patients in both groups showed a mean improvement in BFMDRS of 71.05% with GPiDBS. Meanwhile, patients with secondary dystonia due to previous perinatal insults showed a mean improvement in BFMDRS of 41.9%, with better results in purely dyskinetic patients (mean improvement of 61.2%).

There has been report on improvement of dystonia by MRgFUS (MR guided focused ultrasound therapy) in PTT. The mean improvement of dystonia in TWSTRS at 6 months was significantly improved by 43.4% ($P<0.001$) from baseline. The BFMDRS-Neck scales at 6 months (4.2 ± 2.8) were significantly improved by 38.2% ($P<0.001$) from baseline.[18,19,20] Similarly there was 70.6 % improvement in TWSTRS score in the study from Russia. [21]These results also comparable to our result on PTT lesioning.

We experienced few surgical complications in our patients with pallidotomy. There was a case of early internal capsule infarction following GPi lesioning leading to hemiparesis.[22] There was transient dysarthria in three cases of pallidootmy. Similarly there was hypophonia in one case of PTT lesioning.

Conclusion

Lesioning surgery is rewarding for cervical dystonia. It is less time consuming and cheaper and it should not be abandoned. Both unilateral PTT lesioning or unilateral pallidotomy is equally effective in cervical dystonia. Though some recurrent cases may benefit from DBS(Deep brain stimulation) surgery we believe lesioning surgery is effective with good outcome.

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