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Globus pallidotomy for the management of post-stroke hemichorea: A case report

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Hemichorea (HC) is an involuntary, forcible, rapid, jerky, non-patterned movement of the distal limbs, while hemiballismus (HB) describes a more severe, high-amplitude proximal movement. Hemichorea-hemiballismus (HC-HB): Rare movement disorders are observed with lesions in the contralateral basal ganglia due to different etiologies. Its pathogenesis can originate from infections, immune reactions, metabolic abnormalities, malignancy, neurodegeneration, vascular diseases, or drugs. The most common etiologies include ischemic stroke, hemorrhagic stroke, and autoimmune conditions.[1]

Stroke is the most frequent etiology, with an incidence of around 0.4%-0.5%. Common stroke locations include the striatum, followed by the subthalamic nucleus (STN), a finding that may be attributed to the frequency of strokes in these regions.[2] In most patients, the symptoms self-resolve or improve with or without the use of neuroleptic medications, especially in cases of stroke. Although rare, hyperglycemia is another potential cause of HB-HC syndrome, also known as chorea-hyperglycemia-basal ganglia syndrome (C-H-BG) in this context.

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Although the exact pathophysiology is unknown, one proposed mechanism suggests that hyperglycemia increases the blood viscosity and disrupts the blood-brain barrier (BBB), consequently leading to temporary ischemia of the neurons in the basal ganglia, one of the most susceptible areas of the brain.[3] Another theory suggests that decreased perfusion and ischemia lead to a shift towards anaerobic metabolism and consequently increased metabolism of gamma-aminobutyric acid (GABA), the main inhibitory neurotransmitter of the basal ganglia.[4]

Clinicians use a variety of methods to treat HC, depending on the underlying cause and severity of symptoms. Medical management typically includes medications such as dopamine-depleting agents (e.g., tetrabenazine), antipsychotics (e.g., haloperidol or risperidone), and anticonvulsants (e.g., valproic acid) to help reduce involuntary movements.[5] In cases where HC is caused by metabolic abnormalities, infections, or autoimmune conditions, addressing the root cause – such as controlling blood sugar in diabetes or using immunosuppressants for autoimmune diseases – is essential.[6,7] For patients with persistent or disabling symptoms that do not respond to medications, surgical interventions like deep brain stimulation (DBS) or lesioning procedures such as pallidotomy may be considered, particularly when the condition is related to structural brain lesions. Multidisciplinary care, including physical and occupational therapy, can also help improve function and quality of life (QOL).[8]

Pallidotomy, a neurosurgical intervention, involves the targeted lesioning of the globus pallidus (GP), a basal ganglia structure critically involved in motor control and the modulation of involuntary movements.[9] The disruption of pathological neural circuitry via this procedure aims to ameliorate HC symptoms. Pallidotomy may be indicated as a treatment modality for HC, specifically in instances of significant symptom severity and when pharmacological interventions have demonstrated insufficient efficacy.[10]

Evaluation of movements in chorea conduct using the Abnormal Involuntary Movement Scale (AIMS): AIMS is a clinical tool used to assess and monitor involuntary movements, particularly those associated with antipsychotic medications. It is most commonly applied to detect tardive dyskinesia (TD), a potentially irreversible movement disorder characterized by repetitive, involuntary movements, often affecting the face, lips, tongue, trunk, and extremities. Regular AIMS screening is recommended to ensure early detection and intervention.[11] This study aims to present the efficacy and necessity of pallidotomy in post-stroke patients who have not responded to pharmacological treatment.

A 68-year-old married patient with no baseline disorders other than hypertension (HTN) suffered a cerebrovascular accident (CVA) in 2018. The patient was not taking any medications aside from valsartan 25 mg, administered twice daily (BD), and did not report any previous history of cancer in themselves or any first- or second-degree relatives. Furthermore, there were no notable findings in the social history regarding tobacco or substance use.

Within a few weeks after CVA, involuntary movements had started. Then it spread to the back and legs. HB with thrashing motions, left hand irregular twisting and writhing are not repetitive or rhythmic, Spastic gate, turning to the side and rolling on the floor, were all movments. The overall score of the patient based on the AIMS criteria before surgery was 12, indicating a high severity of symptoms (Video 1).[12]

2018 brain magnetic resonance imaging (MRI) demonstrated an old CVA (Figures 1, 2). Tab tetrabenazine 25 mg and cap amantadine hydrochloride 100 mg did not respond to her. By tab haloperidol 0.5 mg and diphenhydramine 5 ml three times a day (TDS), the movements were completely controlled. In 2022, HC movements returned to their former intensity, along with the additional movements of the neck and jaw. Olanzapine 5 mg BD, memantine 5 mg BD, quetiapine 25 mg daily, tetrabenazine 25 mg, and amantadine 100 mg did not yield any response. It relatively responded to treatment by tab aripiprazole 5 mg 1/2 BD and cap gabapentin 100 mg BD. She got major depressive disorder (MDD) 18 months after CVA (Table 1). Other etiologies for HC, including hyperglycemia, hypoglycemia, or autoimmune disease such as Sjögren's syndrome, multiple sclerosis (MS), and lupus, were excluded by laboratory tests; therefore, the patient was scheduled for surgery.

To prepare her for the pallidotomy, the patient was not intubated; local anesthesia was induced, and the patient underwent awake surgery. A Cosman-Roberts-Wells (CRW) frame was applied, and the stereotactic magnetic resonance (MR) images were obtained using the standard protocol for pallidotomy.

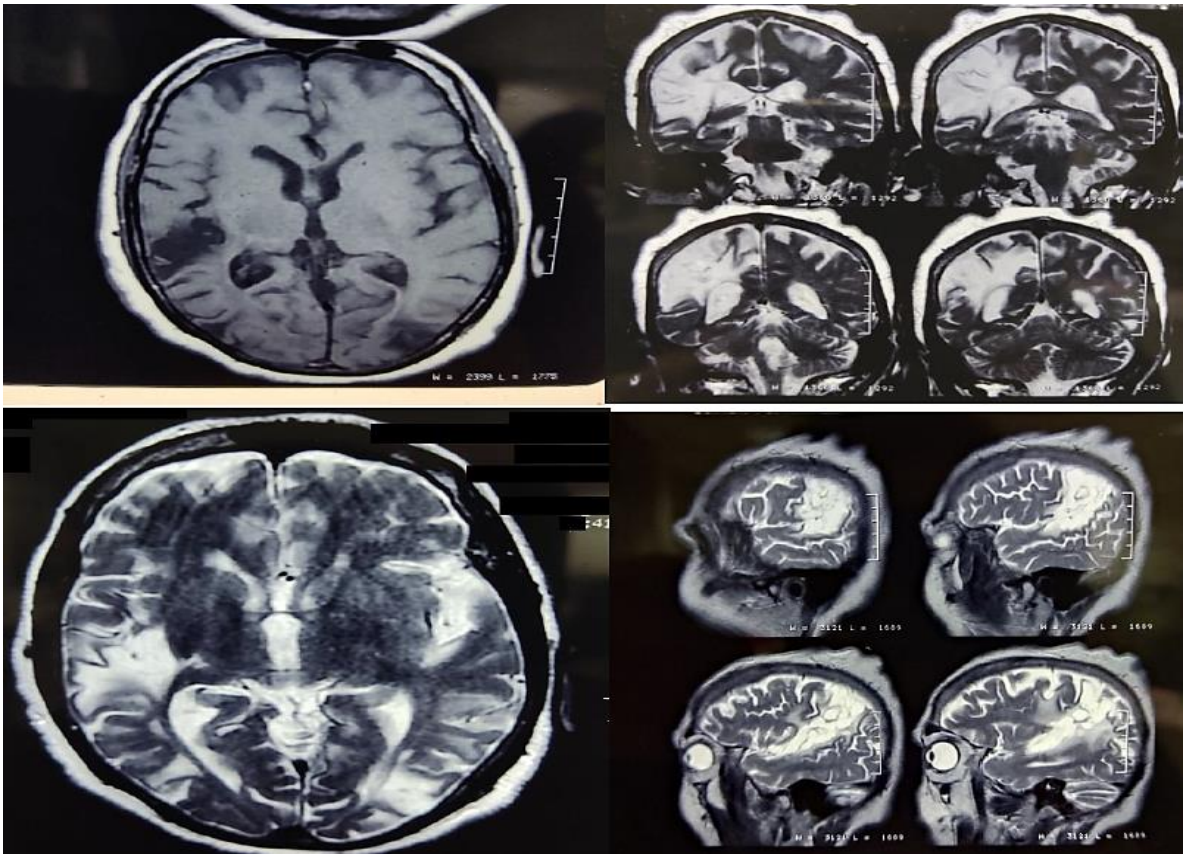

Figure 1. Pre-operative magnetic resonance imaging (MRI): T1-weighted axial view, T2-weighted axial view, T2-weighted coronal view, T2-weighted sagittal view

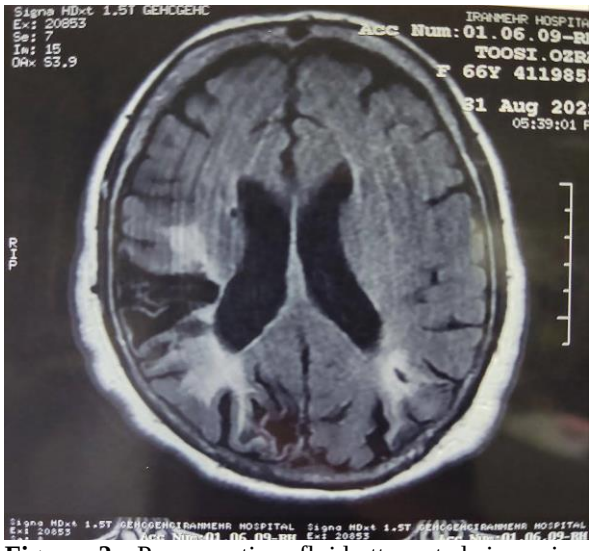

Figure 2. Pre-operative fluid-attenuated inversion recovery (FLAIR) magnetic resonance imaging (MRI), axial view

Axial T1-weighted, axial inversion-recovery, and coronal fast spin echo (FSE) MR images were obtained. The GP, internal capsule, and optic tract were identified on both sides. Subsequently, the stereotactic coordinates for the target point were calculated. The target was chosen at 19 mm to the right of the midline, 2 mm anterior to the mid commissural point, and 6 mm inferior to the plane of the anterior commissure-posterior commissure (AC-PC). After targeting was completed, the cannula was inserted through the twist-drill opening in the right pre-coronal region parallel to the sagittal plane and at a 65° angle to the axial plane. After completion of the micro recording, the standard method of radiofrequency thermocoagulation was used after heating the tip of the probe to 84 °C for 60 seconds. A standard 1.1-mm-diameter probe with a 2-mm bare tip was used for the pallidotomy; subsequently, the electrode and cannula were removed. The surgery result was perfect; the patient did not report any adverse events or complications, and we did not see anyone. She was treated successfully. This patient was monitored and followed up for 12 months, during which comprehensive neurological examinations were conducted at intervals of 1, 3, 6, and 12 months. In the follow-up assessments, the score achieved by the patient according to AIMS criteria was 2, which reflects a significant improvement in the patient's condition.

Table 1. Timeline of the patient's treatments and interventions

CVA	February 2018
Start of movement disorder	March 2018
Diagnosis of major depressive disorder	May 2019
Pharmacological treatment	March 2018 to May 2022
Globus pallidotomy	May 2022
One-month postsurgical follow-up	June 2023
Three-month postsurgical follow-up	August 2023
Six-month postsurgical follow-up	November 2023
One-year postsurgical follow-up	May 2023

CVA: Cerebrovascular accident

Extra body movements were controlled and treated (Video 1). A post-operation computed tomography (CT) scan shows the lesion in the right GP internus (GPi) (Figure 3). The current study has been reported in line with the CAse REport (CARE) criteria.

This case report discusses the successful treatment of HC in a patient post-CVA through globus pallidotomy. The procedure involved targeted lesioning of the GP, aiming to alleviate the abnormal involuntary movements associated with HC. The report delves into the patient's preoperative assessment, surgical technique, and postoperative outcomes, highlighting improved motor function and QOL. This intervention presents a promising avenue for managing HC following a CVA, shedding light on the potential efficacy of globus pallidotomy in addressing post-stroke movement disorders.

The most prevalent etiology of HC is an acute ischemic or hemorrhagic stroke. Hyperglycemic HC is also noted. Additional etiological factors include hypoglycemia, Sydenham's chorea, uremia, underlying genetic disorders, autoimmune conditions such as lupus, MS, or Sjögren's syndrome, as well as endocrine or metabolic disorders like hyperthyroidism or kernicterus. Infections or complications from diseases, such as rheumatic fever leading to Sydenham's chorea, may also contribute. Furthermore, the presence of a brain tumor adjacent to the basal ganglia can be implicated. For this patient, firstly, we ruled out other etiologies and, in the last step, chose globus pallidotomy for treatment.

Globus pallidotomy involves the precise lesioning of the GP, a critical component of the basal ganglia involved in motor control. This targeted approach disrupts aberrant neural signaling responsible for HC, providing a direct avenue for symptom relief.[13] Successful cases often report significant improvements in motor function post-globus pallidotomy, allowing for the restoration of more controlled and coordinated movements. Studies by Loher et al.[14] and Albanese et al.[15] have highlighted substantial improvements in motor function following globus pallidotomy interventions.

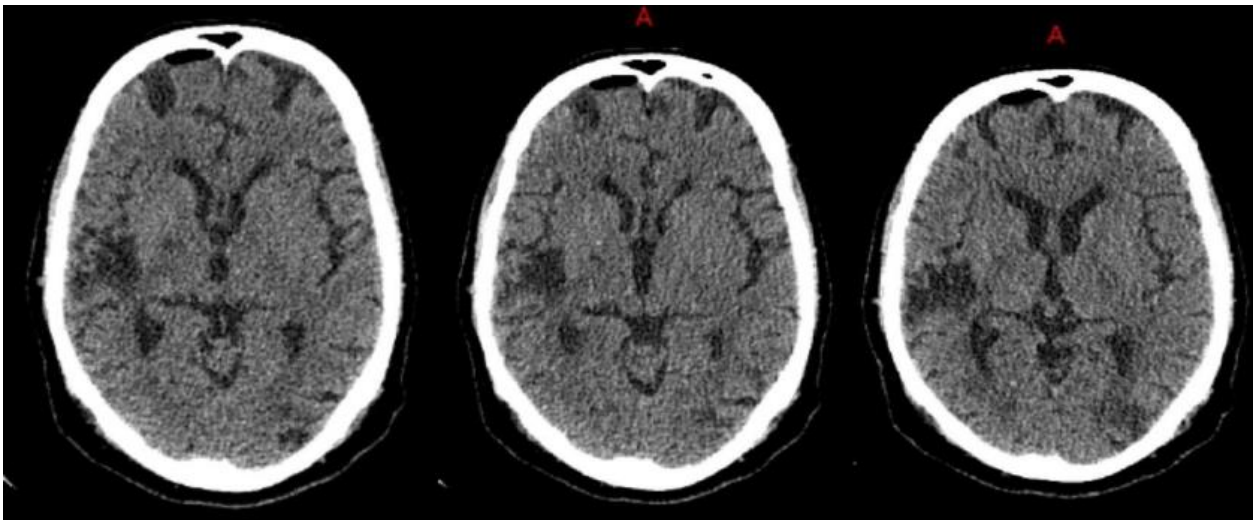

Figure 3. Post-operative computed tomography (CT)-scan; lesion location is highlighted

HC, impacting a patient's daily activities and overall QOL, can be significantly alleviated by globus pallidotomy, enhancing the ability to perform daily tasks independently. Bhatia and Marsden demonstrated improvements in QOL metrics following globus pallidotomy for HC.[16] As with any neurosurgical procedure, globus pallidotomy carries inherent risks, including bleeding, infection, and neurological deficits. The delicate nature of the brain and its intricate connections necessitate a thorough risk-benefit analysis before opting for such interventions. The outcomes of globus pallidotomy can vary among individuals, influenced by factors like lesion size, precise targeting, and individual patient characteristics. Kleiner-Fisman et al. emphasized the need for comprehensive preoperative assessments to optimize patient selection and outcomes.[13] Long-term follow-up data on the efficacy and safety of globus pallidotomy for HC post-CVA are relatively limited, although some studies indicate that outcomes and QOL in patients are favorable as same as we see in our case.[17] Another study by Tatang and Inggas presented a case report of pallidotomy for treating chorea with acceptable outcomes,[18] which was compatible with our findings. Comprehensive studies with extended follow-up periods are crucial to assess the durability of symptom relief and identify potential late complications. Ongoing research by Vidailhet et al. explores the long-term outcomes of globus pallidotomy, shedding light on sustained benefits and potential challenges.[19] Various studies have explored pharmacological treatments for HC post-CVA, including dopamine receptor blockers. While these medications may provide symptomatic relief, their efficacy can be variable, and they may be associated with side effects. Studies suggested that surgical interventions, including globus pallidotomy, may offer more consistent and significant improvements in motor function compared to pharmacological approaches.[5] DBS, another neurosurgical intervention used in the management of movement disorders, involves the implantation of electrodes in specific brain regions to modulate activity through electrical stimulation. Comparative studies between globus pallidotomy and DBS highlight similar efficacy in improving motor symptoms, with the choice often influenced by factors such as patient preference and surgical risks.[20] Ostrem et al. suggested that while both interventions showed promise, individual patient characteristics played a crucial role in determining the most appropriate treatment approach.[21]

We could not follow the patient for more than one year because she moved to another city.

In conclusion, globus pallidotomy emerges as a promising and effective option for treating HC in post-CVA patients, with advantages in targeted lesioning, motor function improvement, and enhanced QOL. However, careful consideration of potential disadvantages, including surgical risks and variable outcomes, is crucial. Comparative analyses with alternative treatments underscore the need for personalized approaches, emphasizing the importance of patient-specific factors in treatment decision-making. Continued research, such as ongoing studies on long-term outcomes, will contribute to a more comprehensive understanding of the efficacy and safety of globus pallidotomy in this context.

Conflict of Interests

The authors declare no conflict of interest in this study.

Acknowledgments

Considering the national committee for ethics in biomedical research laws, it is not necessary to get the ethical approval code for case reports; only patients' consent is enough.

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