Silent Arteriovenous Malformation, Loud Movements: A Dyskinetic Puzzle Resolved by Staged Craniotomy and Immediate Globus Pallidus Internus Deep Brain Stimulation

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Date of Submission: 17th January 2026 Date of Acceptance: 7th March 2026 Date of Publication: 15th March2026

Abstract

Background: Tardive dyskinesia is a debilitating hyperkinetic movement disorder characterized by repetitive involuntary movements involving the orofacial musculature. The disorder most commonly arises following exposure to dopamine receptor–blocking medications and reflects dysfunction within cortico-basal ganglia-thalamocortical motor circuits[1]. Structural brain lesions may further destabilize motor network function and contribute to abnormal movement patterns. Cerebral arteriovenous malformations (AVMs) are congenital vascular anomalies that typically present with intracranial hemorrhage, seizures, headaches, or progressive neurological deficits[2]. Movement disorders associated with AVMs are rare.

Case Description: We report a case of severe refractory oro-buccal dyskinesia in a patient with a cerebral AVM and seizure disorder. Neuroimaging demonstrated a left posterior temporal arteriovenous malformation located near the temporoparietal junction. Despite withdrawal of the offending medication and trials of tetrabenazine and botulinum toxin injections, the dyskinesia persisted. The patient underwent microsurgical AVM excision, which resulted in angiographic cure and seizure control but did not resolve the abnormal movements. Because of persistent disabling dyskinesia, the patient subsequently underwent bilateral globus pallidus internus deep brain stimulation (GPi-DBS). Intraoperative microelectrode recordings confirmed appropriate pallidal neuronal activity. An implantable pulse generator was implanted during the same procedure and stimulation was activated immediately in the recovery period, resulting in rapid suppression of dyskinetic movements.

Conclusion: This case highlights the complex interaction between structural cerebrovascular pathology and dysfunctional basal ganglia circuitry. Successful treatment required a staged neurosurgical strategy combining vascular and functional neurosurgery. Immediate activation of pallidal stimulation produced dramatic clinical improvement and demonstrates the therapeutic role of neuromodulation in refractory dyskinetic disorders.

Keywords: Arteriovenous malformation ,Deep brain stimulation ,Globus pallidus internus ,Movement disorder ,Tardive dyskinesia

Introduction

Tardive dyskinesia is a chronic hyperkinetic movement disorder characterized by repetitive involuntary movements involving the lips, tongue, jaw, and facial musculature. The disorder most commonly develops following prolonged exposure to dopamine receptor–blocking medications and reflects dysfunction within cortico-basal ganglia-thalamocortical motor circuits[1]. Although the clinical syndrome has been well described, the underlying pathophysiology remains incompletely understood. Proposed mechanisms include dopamine receptor hypersensitivity, impairment of inhibitory gamma-aminobutyric acid signalling, oxidative neuronal injury, and maladaptive synaptic plasticity within basal ganglia pathways[3].

Structural lesions of the central nervous system may also influence motor behaviour by altering distributed neural networks. Lesions involving cortical motor regions, thalamic relay nuclei, or basal ganglia structures can disrupt the balance of excitatory and inhibitory signalling required for coordinated voluntary movement[3]. Even lesions outside the basal ganglia may affect motor circuitry through network-level interactions.

Cerebral arteriovenous malformations are congenital vascular anomalies characterized by direct arterial-to-venous shunting without an intervening capillary bed. AVMs most commonly present with intracranial hemorrhage, seizures, headaches, or progressive neurological deficits[2]. Although uncommon, movement disorders associated with vascular lesions have been reported, likely due to mechanisms such as cortical irritation, hemodynamic steal, or disruption of motor pathways[4].

Access this article online

Website: https://www.nepjol.info/index.php/NJN

DOI: https://doi.org/10.3126/njn.v23i1.91823

HOW TO CITE
Gannison, L., & Muthusamy, K. A. Silent Arteriovenous Malformation, Loud Movements: A Dyskinetic Puzzle Resolved by Staged Craniotomy and Immediate Globus Pallidus Internus Deep Brain Stimulation. NJNS. 2026;23(1):48-51

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ISSN: 1813-1948 (Print), 1813-1956 (Online)

Deep brain stimulation (DBS) has emerged as an effective therapy for medically refractory movement disorders. In particular, stimulation of the globus pallidus internus (GPi) has demonstrated significant benefit in dystonia and tardive dyskinesia by modulating abnormal basal ganglia output and restoring physiological thalamocortical motor signalling[5–8].

We report a rare case of severe refractory oro-buccal dyskinesia associated with a cerebral AVM. Successful treatment required a staged neurosurgical approach combining microsurgical AVM resection and bilateral GPi deep brain stimulation with immediate postoperative activation.

Case Presentation

The patient first presented in 2013 with generalized tonic–clonic seizures. Magnetic resonance imaging and digital subtraction angiography demonstrated a left posterior temporal arteriovenous malformation located near the temporoparietal junction characterized by a compact nidus and early venous drainage (Figure 1).

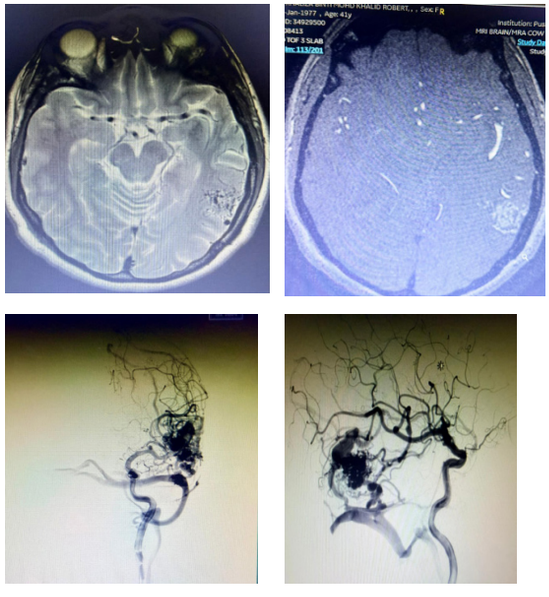

Figure1: Preoperative MRI and DSA demonstrating a left posterior temporal AVM near the temporoparietal junction.

The patient was initially started on levetiracetam for seizure control but subsequently defaulted neurosurgical follow-up and definitive treatment of the AVM was not pursued.

Several years later the patient developed recurrent seizures and antiepileptic therapy was restarted. Neurosurgical evaluation was again recommended.

In 2020 the patient sought psychiatric consultation for progressive jaw clenching and involuntary facial movements interfering with speech and oral intake. Clonazepam was prescribed for symptomatic control. Shortly after initiation of this medication the patient developed worsening oro-buccal dyskinesia characterized by repetitive jaw contractions and facial movements.

Clonazepam was discontinued due to suspicion of medication-induced dyskinesia. However, the abnormal movements persisted. Trials of tetrabenazine and botulinum toxin injections failed to produce meaningful improvement.

The case was discussed in a multidisciplinary team meeting involving neurosurgery, neurology, and psychiatry, and a staged surgical strategy was planned.

Stage 1 – Microsurgical AVM Resection

The patient underwent microsurgical excision of the left posterior temporal AVM through a tailored temporoparietal craniotomy.

Under the operating microscope, circumferential dissection of the malformation was performed. Arterial feeders were sequentially identified and coagulated while preserving normal en-passage vessels. Progressive devascularization of the nidus was achieved before division of the draining vein and complete removal of the malformation.

Postoperative MRI confirmed complete resection of the AVM with no residual nidus identified (Figure 2).

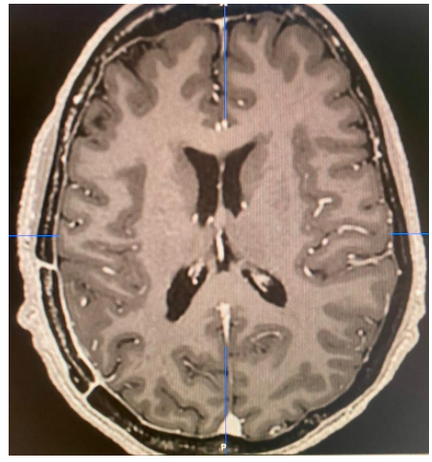

Figure2: Postoperative MRI demonstrating complete resection of the AVM with no residual nidus.

Following surgery the patient achieved complete seizure control, but the dyskinetic movements persisted.

Stage 2 – Deep Brain Stimulation

Because of persistent disabling dyskinesia, the patient underwent bilateral GPi deep brain stimulation.

Preoperative stereotactic planning was performed using high-resolution MRI to identify the GPi targets (Figure 3).

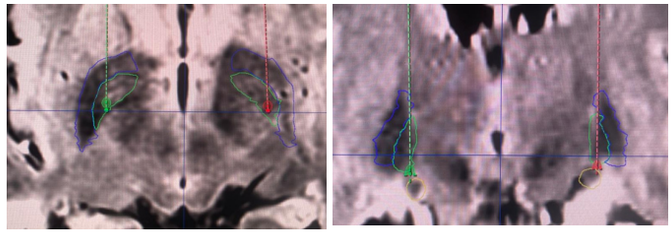

Figure3:Preoperative stereotactic planning demonstrating bilateral GPi targets.

Advanced segmentation techniques were used to reconstruct the globus pallidus externus, globus pallidus internus, and optic tract to optimize trajectory planning and avoid injury to adjacent structures (Figure 4).

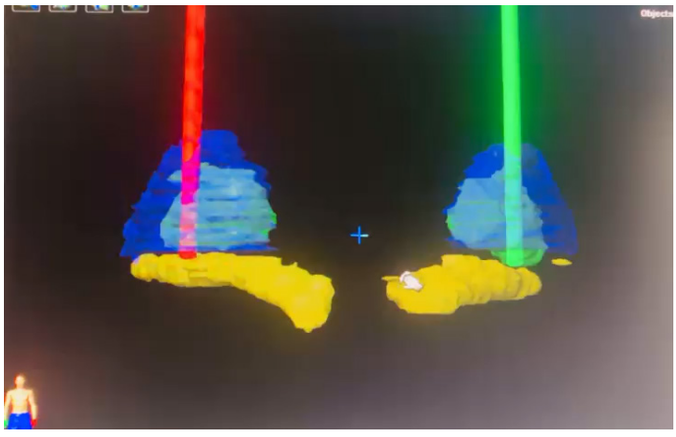

Figure4:Three-dimensional reconstruction showing segmentation of GPe, GPi, and optic tract.

Bilateral stereotactic electrode implantation was performed. Microelectrode recordings (MER) were used intraoperatively to confirm characteristic pallidal neuronal firing patterns, allowing refinement of target localization.

Permanent DBS leads were implanted bilaterally and secured to the skull. Extension leads were tunneled subcutaneously to the infraclavicular region where an implantable pulse generator (IPG) was implanted.

Postoperative CT–MRI fusion imaging confirmed accurate bilateral GPi lead placement (Figure 5).

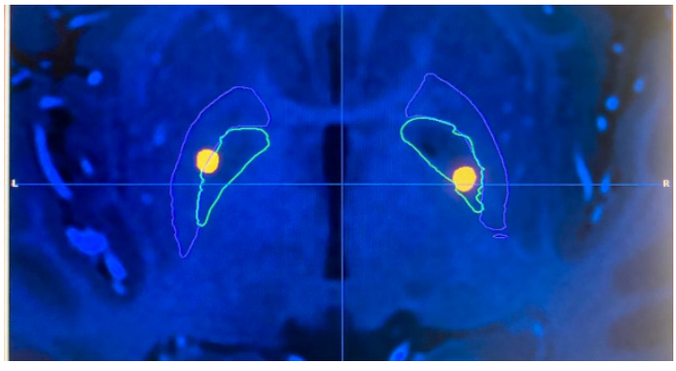

Figure 5: Postoperative CT–MRI fusion demonstrating accurate bilateral GPi DBS lead placement.

Discussion

To our knowledge, reports describing staged microsurgical AVM resection followed by pallidal deep brain stimulation with immediate postoperative activation for refractory dyskinesia are exceedingly rare. Tardive dyskinesia represents a complex disorder involving abnormal signalling within cortico-basal ganglia-thalamocortical motor circuits[13]. Although classically associated with dopamine receptor blockade, structural brain lesions may also contribute to abnormal motor network activity.

In this case, the development of dyskinesia following clonazepam exposure is unusual, suggesting that medication exposure may have acted as a trigger within a neural network already vulnerable due to chronic structural pathology. The presence of a cerebral AVM may alter cortical excitability or disrupt distributed motor pathways.

Movement disorders associated with structural brain lesions have been described but remain uncommon[4]. Proposed mechanisms include cortical irritation, venous hypertension, and hemodynamic disturbances affecting motor circuits.

The most instructive feature of this case is the staged neurosurgical management. Microsurgical excision of the AVM eliminated the structural lesion and resulted in seizure freedom. However, persistence of dyskinesia following AVM removal suggested that abnormal motor circuitry had become independent of the original lesion.

Deep brain stimulation of the globus pallidus internus has demonstrated significant therapeutic benefit in dystonia and tardive dyskinesia[5–8]. Pallidal stimulation modulates abnormal basal ganglia output and restores physiological thalamocortical signalling.

The immediate activation of DBS in this case produced rapid suppression of dyskinetic movements, reinforcing the central role of pallidal circuitry in the generation of the dyskinetic phenotype.

This case therefore illustrates how vascular neurosurgery and functional neurosurgery may be sequentially integrated to address complex neurological disorders involving both structural pathology and dysfunctional motor circuits.

Conclusion

Severe dyskinesia may arise from complex interactions between structural cerebrovascular pathology and dysfunctional basal ganglia circuitry. Successful treatment in this case required a staged neurosurgical approach combining AVM excision and pallidal deep brain stimulation. Immediate activation of stimulation resulted in rapid resolution of disabling dyskinesia.

ACKNOWLEDGEMENT

We Thank our Patient for trust on our team

FUNDING: No fund associated to this case report.

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