Post-Stroke Stereotypies in Aneurysmal Subarachnoid Haemorrhage with Thalamic Infarct – A Case Report

Stereotypies are involuntary, coordinated, repetitive, patterned, rhythmic, and seemingly purposeless movements.[1] Only a few cases of stroke-induced stereotypies have been reported. Infarcts in different brain regions have been implicated in this disorder, such as the lenticulostriatal, thalamic, midbrain, and left middle cerebral artery territory.[2,3] We describe here a patient who developed delayed stroke stereotypy consequent to a contralateral thalamic infarct.

A 50-year-old lady presented to the emergency with a history of sudden-onset, severe headaches for 15 days. The headache had further aggravated and was associated with vomiting at the time of presentation. She had drooping of the right eyelid for 5 days prior to the presentation. There was no loss of consciousness or seizures. No comorbid illnesses or similar past episodes were noted. On examination, her GCS was E2V3M5 at presentation. She had a dilated and non-reacting right pupil with ptosis – consistent with a right oculomotor nerve palsy. There were no other neurological deficits.

Non-contrast CT head showed thick diffuse subarachnoid hemorrhage (SAH), primarily in the right Sylvian fissure and basal cisterns, with extension to the fourth ventricle. CT angiogram showed a 10x2 mm saccular aneurysm at the origin of the right posterior communicating artery, (modified Fisher grade IV). She underwent a right pterional craniotomy and clipping of an aneurysm on the same day [Figure 1f]. Postoperatively, noradrenaline infusion and supplementary intravenous fluids were continued to maintain hypertension and dilutional hypervolemia and to prevent vasospasm, for up to 5 days after surgery.

On postoperative day 6, she was found to have weakness in the left upper limb (MRC grade 2/5). CT head did not show any obvious infarct, but suspecting vasospasm and delayed ischaemic neurological deficit (DIND), MR imaging, and digital subtraction angiogram (DSA) were also performed [Figure 1, a-c]. These investigations revealed cerebral ischemia and an evolving infarct in the right internal carotid artery (ICA) territory [Figure 1, d]. Intra-arterial nimodipine was injected for managing the vasospasm, followed by an infusion of intravenous milrinone and noradrenaline. Her left upper limb weakness improved, and a repeat MRI done 10 days later showed that the previously noted ischaemic area had resolved.

Seventeen days after the surgery, she developed abnormal hyperkinetic movements of the left upper and lower limbs – which were regular, rhythmic, partially suppressible, purposeless, and would disappear in sleep [Video 1]. A diagnosis of "post-stroke stereotypy" was made. MRI brain repeated at this point showed an evolving right thalamic infarct involving the posterior dorsal and ventral posterior nuclei (VPN) of the thalamus with a resolution of the previous territory of ischemia [Figure 1e]. Oral nimodipine was restarted for the vasospasm and low-dose haloperidol (0.25 mg twice a day) was added. The stereotypical limb movements subsided

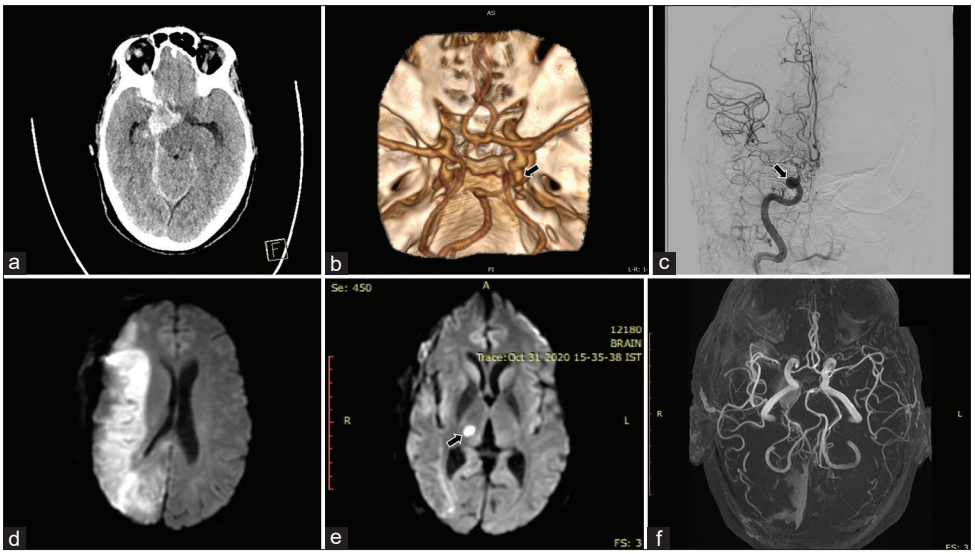

Figure 1: (a) Non-contrast CT head showing thick, diffuse subarachnoid haemorrhage, primarily in right sylvian fissure and basal cisterns (b) Volume-rendering technique 3D reconstruction of the CT angiogram showing a saccular aneurysm (arrow) at the origin of right posterior communicating artery (c) Digital subtraction image (DSA) done preoperatively showing the saccular aneurysm (arrow) at origin of right posterior communicating artery (d) Postoperative MRI diffusion-weighted image (DWI) showing a right internal carotid artery (ICA) territory evolving infarct (e) Postoperative MRI done after 2 weeks showing resolution of the right ICA territory ischaemia, but presence of a right thalamic infarct (arrow) (f) Postoperative MR angiogram image showing obliteration of the aneurysm post-clipping

over the next two weeks, barring a mild residual left-hand grip weakness.

She was followed up in the outpatient department 15 days later – the stereotypical movement had completely disappeared [Video 1]. Mild left upper and lower limb weakness was persistent. Haloperidol was subsequently discontinued.

Cerebrovascular diseases, most commonly strokes, represent a large fraction of the aetiology of movement disorders. Strokes can lead to movement disorders such as hemichorea, hemiballismus, dystonia, tremors, myoclonus, asterixis, stereotypies, and vascular parkinsonism. These mainly affect the deep structures of the brain – the basal ganglia (44%) and thalamus (37%). They are usually unilateral and clinically manifest contralateral to the brain lesion.[3] Stroke stereotypies are "involuntary, coordinated, repetitive, patterned, and rhythmic, seemingly purposeless movements".[4] These abnormal movements can occur immediately following the stroke or can be delayed and progressive. A, younger age at the onset of injury has been seen to be associated with longer latency, and a greater tendency to develop generalized rather than focal or segmental manifestations.[5]

Stroke stereotypies (both ischemic and hemorrhagic), though rare, are most frequently associated with strokes involving the thalamus, midbrain, or putamen.[3,5,6] Laplane *et al*.[7] reported two cases of stereotypy with bilateral basal ganglia lesions while Lee *et al*.[8] reported a case following cerebellar infarction. In the Lausanne Stroke Registry, only 29 of the 2,500 stroke patients (1%) developed post-stroke hyperkinetic abnormal movements, and only two (<1%) patients developed stereotypies.[9]

Although the exact pathophysiology of post-stroke stereotypic movements is still unclear, it is postulated that the cortico-striatal-thalamo-cortical and cerebello-thalamic pathways, required to inhibit involuntary movements while performing voluntary actions, are disrupted. Disruption of these pathways can give rise to abnormal involuntary movements including stereotypies.[2,9]

The phenomenology of post-stroke stereotypies can be simple or complex. Continuous tapping of the contralateral hand secondary to unilateral thalamic infarct is an example of simple stereotypy; while repeated pirouetting while walking, or repeated ritualistic movements like hand-clapping, flailing arms, whispering, protruding the tongue, etc., are examples of complex ones. Ipsilateral instinctive grasp and stereotyped involuntary movements like groping or picking have been described with right frontal lobe infarction. Tics are the closed mimics of stereotypies, although, unlike stereotypies which are rhythmic, repetitive tics are sudden, brief, rapid, and non-rhythmic.[3]

The paucity of published literature and inherent biases of case reports have made it difficult to evolve any definitive treatment guideline for stroke stereotypies. Tetrabenazine, the most effective drug for tardive stereotypies, has not been evaluated in patients with post-stroke stereotypies. Clonazepam can be used but stereotypy is usually refractory to benzodiazepines, amantadine, and anticholinergics.[6] Our patient showed a remarkable response to haloperidol used in low doses and for a short duration.

To conclude, post-stroke stereotypy can be a delayed manifestation of cerebral ischemia involving the thalamus and basal ganglia as

in our case with DIND after aneurysmal SAH. Short-duration haloperidol may be a treatment option in post-stroke stereotypy.

Declaration of patient consent

Written Informed Consent was taken from the patient and attendants for publication of clinical images and videos.

Abbreviations

GCS – Glasgow Coma Scale
CT – Computed Tomography scan
MRI – Magnetic Resonance Imaging
MRC – Medical Research Council scale.

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Conflicts of interest

There are no conflicts of interest.

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