Case Report

Subarachnoid Hemorrhage and Internal Carotid Artery Dissection and Occlusion Following Self-Enucleation

Abstract

Self-enucleation is an uncommon type of major self-injury, which may lead to severe neurological deficits and life-threatening complications, such as subarachnoid hemorrhage (SAH) and internal carotid artery (ICA) dissection and occlusion. Our patient is a 53-year-old man with a history of bipolar disorder and schizophrenia who presented with SAH, intraventricular hemorrhage, ICA dissection and occlusion, and right cerebral infarct following self-enucleation. Despite a Glasgow Coma Score of 6 on initial presentation, he improved with conservative management. He achieved a near-complete neurological recovery, with residual left lower extremity weakness and mild confusion. Self-enucleation is a major neurologic, ophthalmologic, and psychiatric emergency with a potential for serious neurological complications and contralateral visual loss. Yet, conservative management may lead to dramatic recovery.

Keywords: *Dissection, internal carotid artery, occlusion, self-enucleation, subarachnoid hemorrhage*

Hadi Joud, Mohammad Hassan A. Noureldine[1], Ivo Peto[2], Jay I. Kumar[2], Jasmina Bajric[3], Siviero Agazzi[2]

University of South Florida Morsani College of Medicine, Departments of [2]Neurosurgery and Brain Repair and [3]Ophthalmology, Tampa General Hospital, University of South Florida Morsani College of Medicine, Tampa, [1]Department of Neurosurgery, Institute for Brain Protection Sciences, Johns Hopkins All Children's Hospital, Johns Hopkins University School of Medicine, Saint Petersburg, Florida, USA

Introduction

Self-enucleation, also known as auto-enucleation or oedipism, is an uncommon type of self-mutilation that is classified as a major self-injury.[1,2] It is more common in men than in women[3,4] and is overwhelmingly associated with acute psychosis due to untreated psychiatric illnesses or drug use.[1,4-6] Self-enucleation involves avulsion of the extraocular muscles, optic nerve, and ophthalmic artery (OA), which may lead to devastating neurovascular complications. These include contralateral visual field deficits due to optic chiasm injury, subarachnoid hemorrhage (SAH), cerebrospinal fluid leak, infection, and internal carotid artery (ICA) dissection.[4]

Only 10 cases of traumatic SAH due to OA avulsion have been reported in the literature,[4,7-9] five of which were reported to be associated with diffuse hemorrhage in the basal cisterns, mimicking the pattern seen in aneurysmal SAH. Given the risk of cerebral vasospasm following SAH, which may lead to ischemic infarcts, these patients require careful monitoring in an intensive care unit.[5] To the best of our knowledge, only one case of ICA dissection after self-enucleation has previously been reported in the literature.[4] We present a unique case of self-enucleation with both SAH as well as ICA dissection and occlusion.

Case Report

The patient is a 53-year-old male with a history of bipolar disorder, schizophrenia, and multiple suicidal attempts who electively stopped his psychiatric medications at 1 month prior to presentation. During an acute psychotic episode, the patient self-enucleated his right eye. He arrived unconscious at the emergency department (ED), and he was intubated. The empty right orbit was washed out and packed with iodoform gauze in the ED, and the patient was then transferred to our facility. Neurological examination revealed a Glasgow Coma Score (GCS) of 6, withdrawal in all extremities except left upper extremity, and positive left corneal, cough, and gag reflexes. Computed tomography (CT) images showed extensive SAH (Fisher grade 4), as well as intraventricular hemorrhage and acute hydrocephalus [Figure 1]. CT

Address for correspondence:
Dr. Siviero Agazzi, Department of Neurosurgery and Brain Repair, Tampa General Hospital, University of South Florida Morsani College of Medicine, Tampa, Florida, USA.
E-mail: sagazzi@usf.edu

How to cite this article: Joud H, Noureldine MH, Peto I, Kumar JI, Bajric J, Agazzi S. Subarachnoid hemorrhage and internal carotid artery dissection and occlusion following self-enucleation. Asian J Neurosurg 2020;15:1050-4.

Submitted: 28-Apr-2020 **Revised:** 14-Jun-2020
Accepted: 10-Aug-2020 **Published:** 19-Oct-2020

Access this article online

Website: www.asianjns.org

DOI: 10.4103/ajns.AJNS_183_20

Quick Response Code:

angiography and digital subtraction angiography (DSA) revealed occlusion of the right ICA [Figure 2]. The gauze in the right orbit was gently removed with irrigation and no bleeding was encountered. A plastic conformer was placed inside the eyelids to maintain the socket space and allow the socket to granulate in, and antibiotic eye ointment was applied to prevent infection.

Per discussion with the family, the patient was initially managed conservatively in the neurointensive care unit. His GCS, however, improved, and he was extubated on posttrauma day (PTD) 2, with a GCS of 13. Mental status then declined overnight. An external ventricular drain (EVD) was placed, revealing an elevated opening pressure of 30 cmH2O, and resulted in immediate improvement. Neurological examination showed a reactive left pupil and full motor strength in all extremities, except for weakness in the left distal lower extremity that persisted throughout the hospital stay. Magnetic resonance imaging (MRI) demonstrated acute ischemia in the right distal anterior cerebral artery (ACA) and middle cerebral artery (MCA) territories [Figure 3]. Repeat cerebral angiogram confirmed complete occlusion of the right cervical ICA (C1–C7 segments) with robust cross-filling of the right MCA from the anterior communicating and posterior communicating arteries [Figure 4]. Daily transcranial Doppler (TCD) readings showed elevated flow velocities in bilateral ACAs and MCAs. The patient failed multiple EVD clamping trials and had persistent hydrocephalus, necessitating placement of a ventriculoperitoneal shunt. Head CT on PTD 28 showed a hypodensity within the right supplementary motor area [Figure 5], consistent with subacute ischemia seen on prior MRI studies and likely the cause of persistent left lower extremity weakness. During a psychiatric evaluation, the patient attributed his self-enucleation act to command auditory hallucinations and was transferred to an inpatient psychiatric unit for continuing care. At discharge, the patient was alert but confused with a GCS of 14. Motor strength was 4+/5 in the right hemibody, 4/5 in the left upper extremity, and 2/5 proximally, and 0/5 distally in the left lower extremity. The patient was given instructions to follow-up for orbital reconstruction with dermis fat graft.

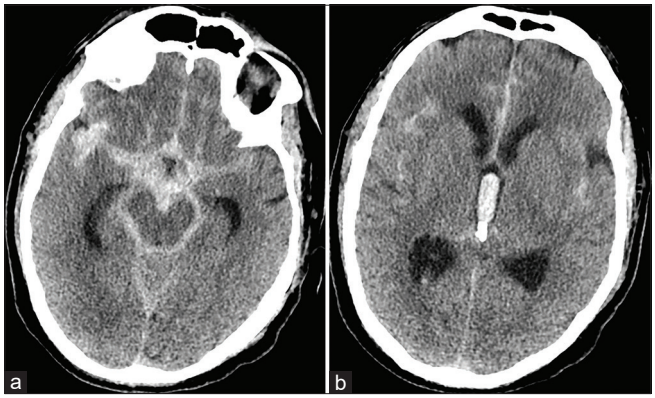

Figure 1: (a) Axial computed tomography head image showing acute diffuse subarachnoid hemorrhage in the frontotemporoparietal and basilar cisterns. (b) Axial computed tomography head images more superiorly showing intraventricular hemorrhage in the 3rd ventricle

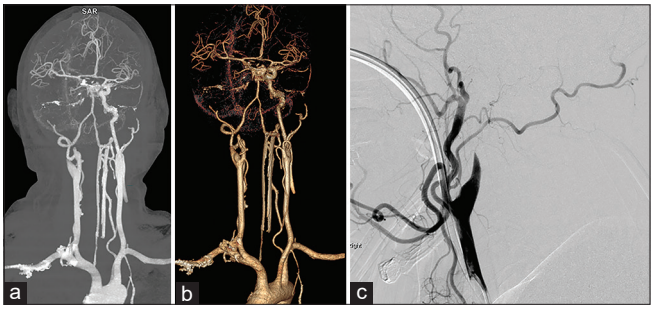

Figure 2: (a) Coronal computed tomography angiography image demonstrating occlusion of the right internal carotid artery just distal to the bifurcation of the right common carotid artery. (b) Coronal three-dimensional reconstruction of the computed tomography angiography image in Figure 2a. (c) Lateral digital subtraction angiography with injection into right common carotid artery demonstrating occlusion of the right internal carotid artery near the bifurcation of the right carotid bifurcation

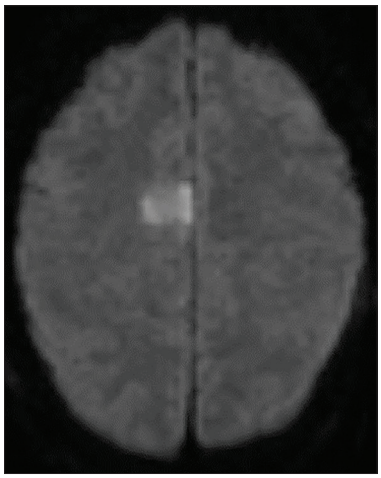

Figure 3: Axial diffusion-weighted imaging image showing a hyperdensity in the right supplementary motor area, indicating an ischemic infarct

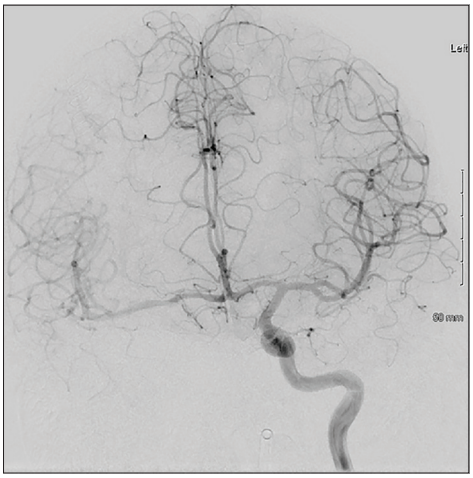

Figure 4: Coronal digital subtraction angiography image with injection into the left internal carotid artery demonstrating filling of the right anterior cerebral and middle cerebral arteries through the anterior communicating artery

Table 1: Summary of literature reporting cases of self-enucleation associated with intracranial complications

Reference (date)	Age	Sex	Laterality	Psychiatric or substance abuse history	Method of self-enucleation	Neurological sequelae	Interventions required for neurological sequelae
Brown (1972)[10]	24	Male	Left	LSD-induced schizophrenic reaction	Own fingers	Contralateral hemianopsia due to optic chiasm damage	None
Coussa *et al.* (2018)[11]	50s	Male	Right	Cocaine	Metallic object	SAH; IVH in lateral, third and fourth ventricles; stroke secondary to vasospasm; cognitive impairment	None
Dilly and Imes (2001)[12]	54	Male	Left	Paranoid schizophrenia; recent discontinuation of medications	Own fingers	Contralateral hemianopsia due to optic chiasm damage	None
Eisenhauer (1985)[13]	33	Female	Right	Schizophrenia	Unknown	Left hemiparesis; aneurysm of ICA at take-off of OA	Surgical clipping of aneurysm
Gauger *et al.* (2015)[4]	37	Male	Left	Schizophrenia; recent discontinuation of medications	Own fingers	SAH in basal cisterns; left ICA dissection and occlusion; RUE and RLE weakness	None
Khan *et al.* (1985)[14]	28	Male	Right	Psychotic depression; alcohol; amphetamines; LSD; marijuana	Own fingers	SAH in the area of the right frontal lobe; LLE numbness and weakness	None
Kotlus and Lo (2007)[5]	31	Female	Left	Alcohol-induced psychosis	Own fingers	Moderate SAH; bilateral stroke and MCA vasospasm; contralateral vision loss	None
Krauss *et al.* (1984)[15]	29	Female	Right	Paranoid schizophrenia	Own fingers	Contralateral hemianopsia due to optic chiasm damage	None
Leibovitch *et al.* (2006)[16]	35	Male	Both	Manic depressive disorder; marijuana; alcohol	Own fingers	Bilateral CSF leaks	Oversowing of annulus of Zinn
Murphy *et al.* (2006)[17]	67	Female	Right	Schizophrenia	Unknown	Diffuse SAH	Unknown
Nabavizadeh *et al.* (2015)[8]	51	Female	Right	"Mental retardation"	Unknown	Diffuse SAH in basal cisterns	None
Rosenthal *et al.* (2011)[18]	46	Male	Left	Schizoaffective disorder; recent discontinuation of medications	Unknown	Bilateral cerebral edema; subdural hematoma; right ICA thrombosis; left cortical stroke; brain death	None
Schargus *et al.* (2009)[19]	84	Male	Left	Advanced dementia; reactive depression	Own fingers	SAH; optic chiasm lesion	None
Shah *et al.* (2017)[20]	46	Male	Both	Schizoaffective disorder	Own fingers	Brain death; SAH with midline shift; subdural hematoma; right ICA thrombosis; left cortical stroke	None
Tabatabaei *et al.* (2011)[21]	36	Male	Right	Ecstasy	Fork	Contralateral hemianopsia due to optic chiasm damage	None
Tuwir *et al.* (2005)[6]	19	Male	Right	Acute psychosis induced by ecstasy; LSD; alcohol	Nail clipper and pliers	Contralateral hemianopsia due to optic chiasm damage	None

CSF – Cerebrospinal fluid; ICA – Internal carotid artery; IVH – Intraventricular hemorrhage; LLE – Left lower extremity; LSD – Lysergic acid diethylamide; MCA – Middle cerebral artery; OA – Ophthalmic artery; RLE – Right lower extremity; RUE – Right upper extremity; SAH – Subarachnoid hemorrhage

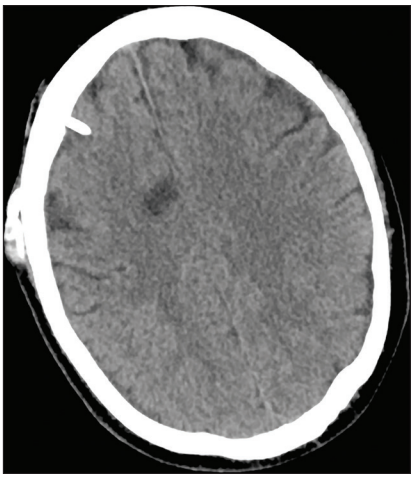

Figure 5: Repeat axial computed tomography head image on posttrauma day 28 showing a hypodensity in the right supplementary motor area, consistent with a subacute infarct

Discussion

We present a unique case of self-enucleation, leading to both SAH and ICA dissection/thrombosis and subsequent coma (GCS 6), followed by a dramatic initial recovery with conservative management only. We also conducted a comprehensive review of the English-language literature for previous cases of self-enucleation that resulted in intracranial complications [Table 1].

We hypothesize that the ICA dissection associated with self-enucleation is a consequence of the traumatic OA avulsion at its take-off from the ICA.[8,9] It leads to hemorrhage in the basilar cisterns mimicking a pattern seen in aneurysmal rupture. Subsequent ICA occlusion results from disruption of the arterial wall, facilitating an acute thrombotic event. Gauger *et al*. postulated that the dissection may result from direct force transmission to the ICA during enucleation.[4] ICA dissection with subsequent occlusion may constitute a life-threatening event in patients without adequate collateral blood flow.[22,23]

Our patient suffered from an ischemic infarct in the right supplementary motor area, which was the likely cause of the left lower extremity weakness. Ischemic infarcts presenting within 2 weeks following SAH are concerning for vasospasm, as in Kotlus and Lo case,[5] which involved bilateral vasospastic strokes following self-enucleation. To assess for vasospasm in our patient, TCD measurements were performed daily and showed persistently elevated flow velocities in multiple arteries, but a subsequent DSA showed no evidence of vasospasm. While TCD is a convenient noninvasive test to screen for cerebral vasospasm,[24,25] DSA remains the gold standard and has a higher diagnostic accuracy.[26] We believe that our patient's infarct was likely not due to vasospasm but rather due to an embolic event precipitated by ICA dissection and occlusion.[27]

A complication commonly reported in previous cases of self-nucleation but not seen in our patient is contralateral temporal field deficit (CTFC).[6,12,15,21,28] CTFC indicates chiasmal injury from avulsion of the optic nerve at its intracranial segment or more than 4 cm proximal to the optic globe and may improve with steroids.[28,29]

Conclusion

Self-enucleation is a dramatic and unfortunate complication of acute psychosis that presents as a neurologic, ophthalmologic, and psychiatric emergency. We reported a unique case of self-enucleation resulting in both SAH and ICA dissection and occlusion that improved initially with conservative management.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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