



# An Eye Opener for a Blinding Disease—Orbital Infarction Syndrome

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## Abstract

### Keywords

- ▶ mechanical thrombectomy
- ▶ OIS
- ▶ orbital infarction syndrome
- ▶ stroke

Orbital infarction syndrome (OIS) is a disease of rare occurrence owing to the rich orbital vascular anastomotic network. We describe a case of a middle-aged female who presented with an acute left middle cerebral artery (MCA) territory infarct with left terminal internal carotid artery and MCA occlusion, underwent emergency mechanical thrombectomy, and developed painful loss of vision shortly after diagnosed as OIS based on clinical and radiological findings. The rarity and severity of OIS, especially in the setting of mechanical thrombectomy, warrant radiologists to be aware of this entity to ensure preventive measures or aid in prompt diagnosis to institute timely treatment.

## Introduction

Orbital infarction syndrome (OIS) is rarely encountered clinically; however, it can be severe enough to cause permanent blindness, and therefore one must be aware of it. Defined as the complete infarction of all intraorbital and intraocular structures, OIS is very rare because of the inherent rich vascular supply of the orbit, principally by the numerous anastomoses between the internal carotid artery (ICA) and external carotid artery (ECA) branches. We described a case of OIS post-routine mechanical thrombectomy, with patent ophthalmic artery (OA) pre- and post-procedure, which makes our case unique and to the best of our knowledge, a first of its kind to be reported in literature.

## Case Report

A 44-year-old female patient with no known comorbidities presented to our emergency department with complaints of

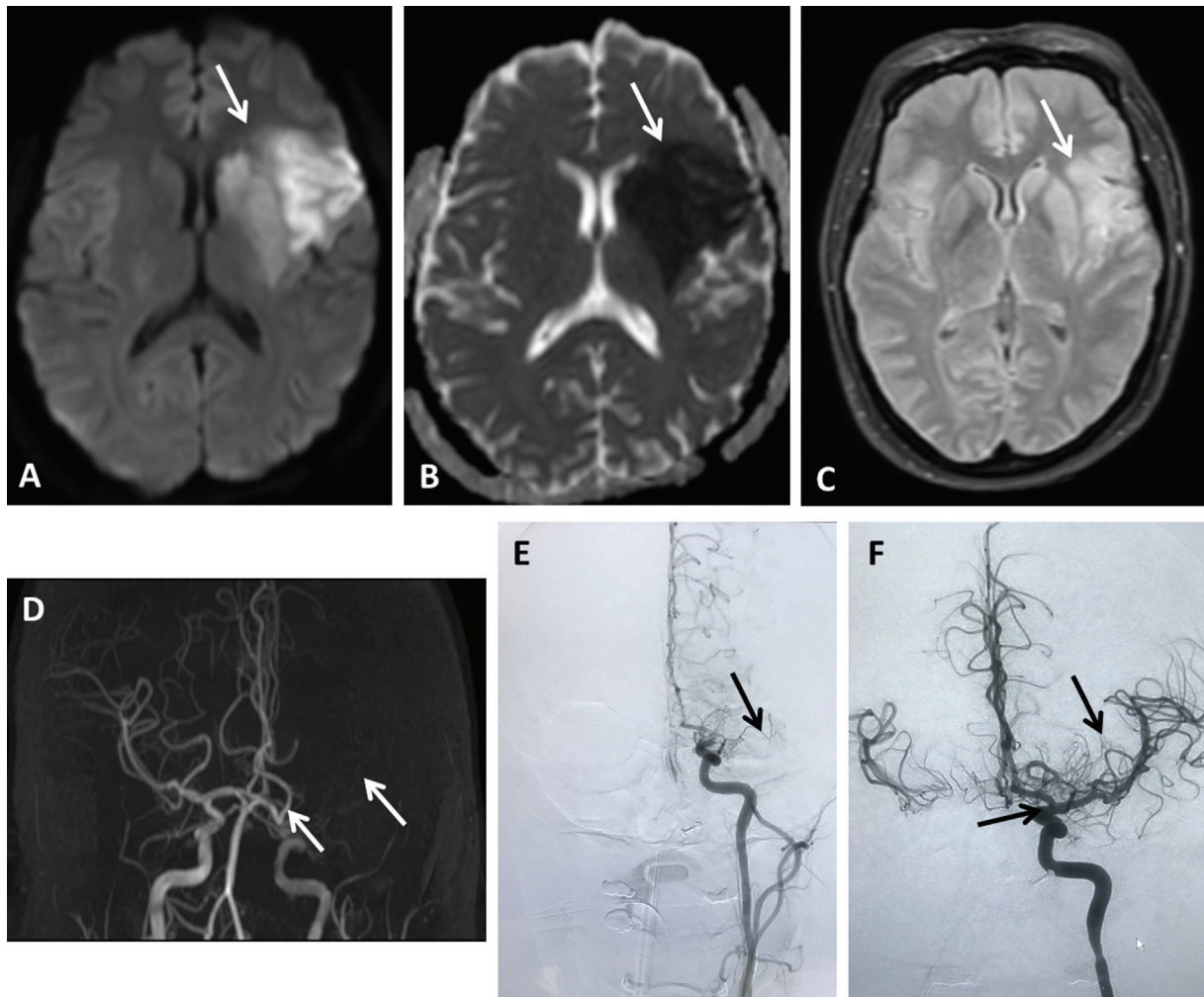
dry cough for the past 2 days, sudden onset right sided weakness, and slurring of speech (~9 hours after stroke symptoms onset) with a score of 14 on National Institutes of Health Stroke Scale. On examination, she was drowsy, disoriented, and not vocalizing (Glasgow Coma Scale [GCS]-E3V1M5).

Postintubation, a magnetic resonance imaging (MRI) brain and angiography was done that showed partial acute left middle cerebral artery (MCA) territory infarct with occlusion of terminal left ICA and left MCA (▶ Fig. 1A–E). Patient underwent an emergency digital subtraction angiography (DSA) and mechanical thrombectomy with door-to-reperfusion time of around 60 minutes and groin-to-reperfusion time of around 25 minutes (▶ Fig. 1F). Workup for stroke in young was normal. Meanwhile, her coronavirus disease 2019 (COVID-19) antigen and reverse-transcription polymerase chain reaction tests returned positive and therefore, etiology of the stroke was attributed to it. Post-procedure, the GCS improved to E3VtM6.

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**Fig. 1** (A, B) Diffusion restriction and (C) subtle fluid-attenuated inversion recovery hyperintense signals involving left neostriatum, anterior and mid-insular cortex and frontal operculum represent acute partial left middle cerebral artery (MCA) territory infarct. (D) Magnetic resonance time-of-flight angiogram showing absent flow-related enhancement of terminal left internal carotid artery (ICA) and left MCA that is confirmed on (E) frontal left ICA shoot on digital subtraction angiography (DSA). (F) Normal flow in left ICA and left MCA post-mechanical thrombectomy as seen on frontal left ICA DSA shoot.

However, patient developed sudden onset swelling of the left eye, a few hours after the procedure. To exclude any procedure-related complications, a contrast computed tomography (CT) brain was done, (5 hours after the first imaging). Left superior ophthalmic vein and cavernous sinus were normal excluding the possibility of a carotid-cavernous fistula (CCF). It revealed axial proptosis, soft tissue thickening in the left preseptal area with extraconal extension external to left lateral rectus muscle, and bulky left extraocular muscles (►Fig. 2A–D). The patient was started on steroids, broad-spectrum topical and intravenous antibiotics, with provisional diagnosis of orbital cellulitis (inflammation/infection related).

A day later with normalized sensorium and complaints loss of vision in the same eye, an evaluation with MRI revealed an increase in extent of the prior noted left orbital changes and T2/short tau inversion recovery (STIR) hyperintensity within the optic nerve, in the intraorbital segment (►Fig. 2E–G). With these imaging findings, a diagnosis of OIS was arrived at.

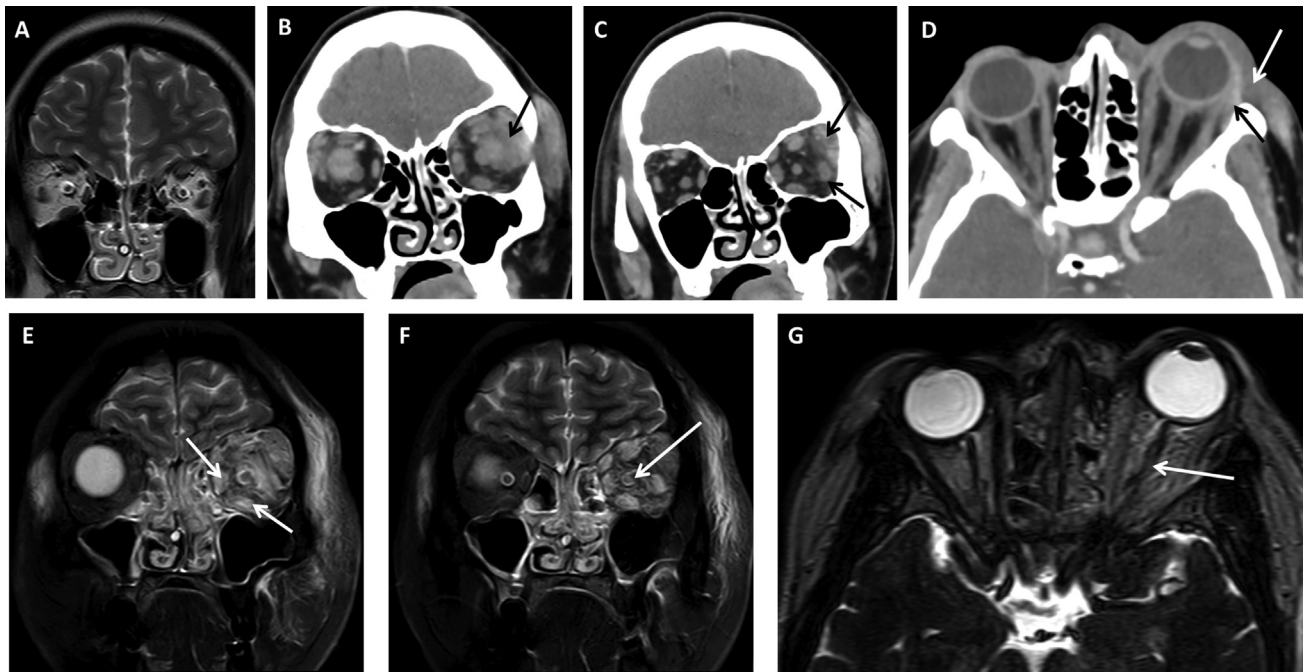
The OA, however, was patent in the initial MRI (►Fig. 3) as well as in the post-procedure CT angiogram (►Fig. 4).

Follow-up MRI brain 1 week later showed stable-sized infarct in the left MCA territory, reduction in the degree of the left orbital changes, and resolution of signal changes within the optic nerve (►Fig. 5). At 2-month follow-up, clinical improvement was seen, with no significant residual weakness; however, the patient had permanent vision loss.

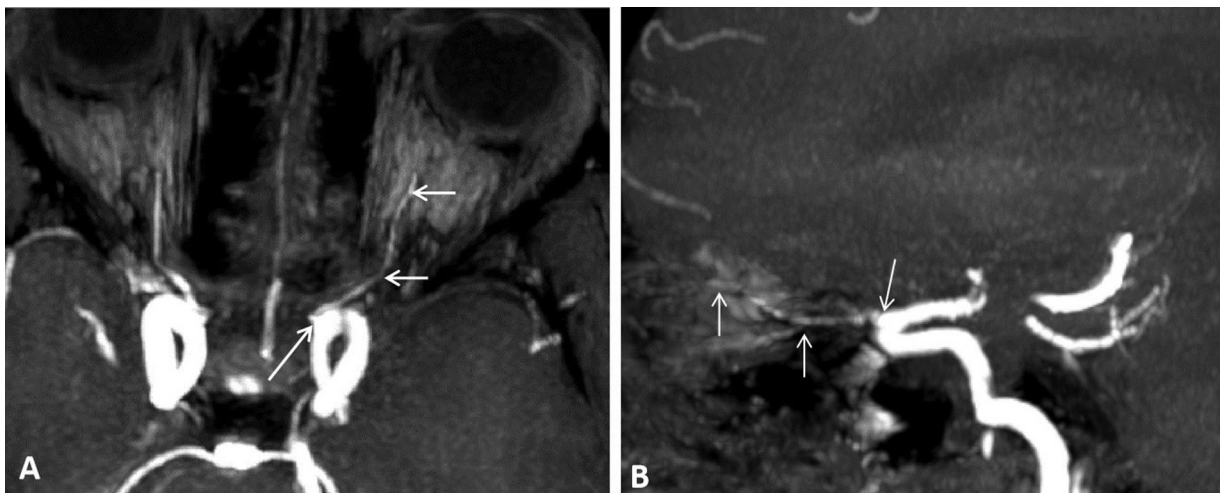
## Discussion

OIS is defined as the complete infarction of all intraorbital and intraocular structures<sup>1</sup> (inclusive of the optic nerve, extraocular muscles and orbital fat), therefore, causing sudden painful vision loss, proptosis, external ophthalmoplegia, chemosis, anterior segment, retinal and choroidal ischemia.

OIS is a very rare disease owing to the rich vascular anastomotic lattice between the branches of the ICA and ECA in the orbit<sup>1–3</sup> which ensure that blood supply to the orbital and ocular structures is maintained even when one of



**Fig. 2** (A) Initial scan with normal orbit, extraocular muscles, and optic nerve bilaterally. (B–D) Computed tomography scan obtained 5 hours later depicting soft tissue thickening in the left preseptal area with extraconal extension external to left lateral rectus muscle. Bulky left extraocular muscles as compared with the right. Axial left eye proptosis. (E–G) Magnetic resonance imaging orbit 1 day later reveals increase in degree of inflammation both in preseptal and postseptal regions of left eye with bulky and short tau inversion recovery (STIR) hyperintense extraocular muscles. (F, G) Interval development of T2/STIR hyperintense signals in the left optic nerve as opposed to a normal right optic nerve. Stable left eye proptosis.



**Fig. 3** Time-of-flight magnetic resonance angiogram: (A) Axial and (B) sagittal maximum-intensity projection images depicting normal flow-related enhancement in the left ophthalmic artery.

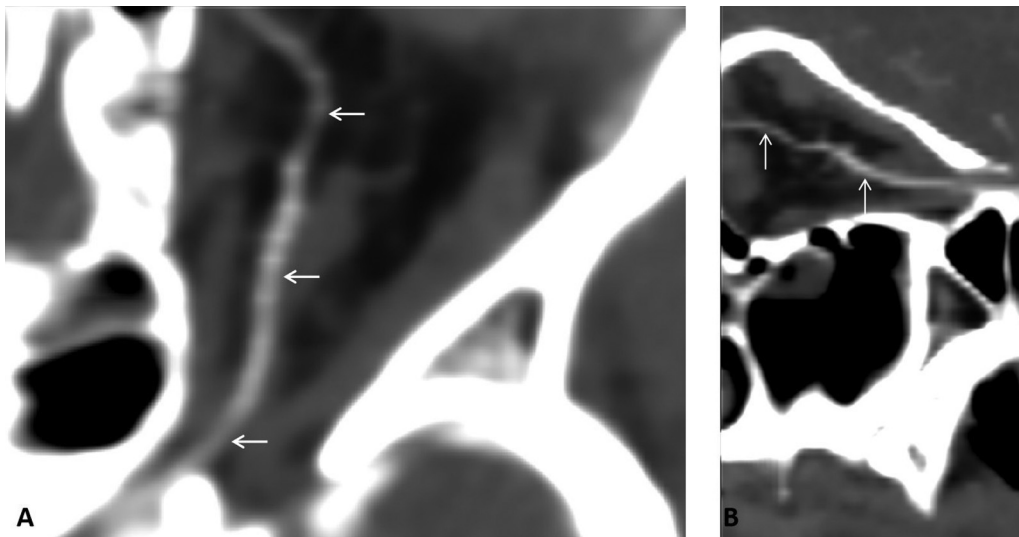
these blood vessels is occluded. A more recent study in children based on DSA throws light on the concept of dominance in the OA based on the direction of flow, either antegrade or retrograde, corresponding to ICA dominance and ECA dominance, respectively, and that this dominance can vary over time.<sup>4</sup> This dynamic dominance is also plausible in adults further substantiating the rarity of occurrence of OIS.

OIS has been reported in literature occurring secondary to many causes like mucormycosis,<sup>1</sup> vasculitis,<sup>5</sup> carotid dissection,<sup>6</sup> and intracranial aneurysmal surgeries<sup>7</sup> to name a few.

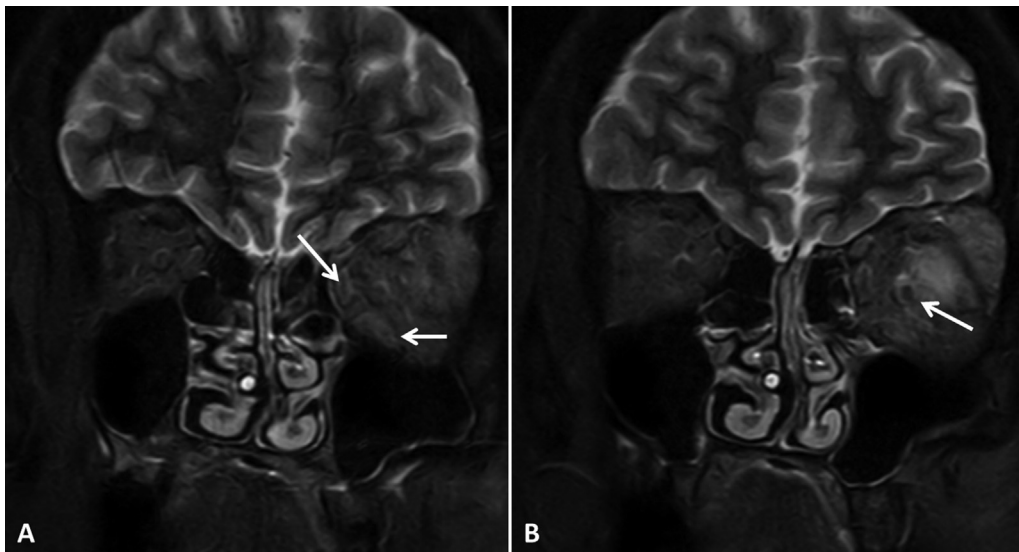
We describe a case of OIS here as a rare complication of mechanical thrombectomy.

OIS as a complication of mechanical thrombectomy has only recently been recognized and reported in literature. The common clinical picture of the cases published<sup>2,3,8-10</sup> is of a patient with acute MCA territory infarct with occlusion of terminal ICA and MCA with variable occlusion of the proximal ICA, subjected to mechanical thrombectomy, who subsequently developed painful loss of vision, eventually diagnosed as OIS. In only one<sup>3</sup> of the cases, the occlusion was seen covering the origin of the OA. In the rest of the cases,





**Fig. 4** Computed tomography angiogram: Curved planar reformation images in (A) axial and (B) sagittal planes depicting normal post-contrast enhancement of the left ophthalmic artery, indicating patency.



**Fig. 5** Follow-up magnetic resonance imaging orbit 1 week later (A) Decrease in degree of inflammation in preseptal and postseptal regions of left eye with reduction in the short tau inversion recovery (STIR) hyperintensity of extraocular muscles (arrows). (B) Interval resolution of STIR hyperintensity within the left optic nerve (arrow) as compared with previous scan.

the OA was patent pre-procedure and only after single or multiple passes for clot retrieval was new occlusion or proximal sluggish flow within the OA detected. Only in one case, the sheath was placed proximal to the carotid bifurcation to the tortuosity of the artery, leading to embolism in both OA and ECA branches.<sup>8</sup>

In a recent retrospective study, the incidence of exophthalmos, all attributed to orbital infarction syndrome post endovascular thrombectomy was 2.4%, with the underlying mechanism postulated to be either due to orbital ischemia from hypoperfusion or distal emboli.<sup>11</sup>

Another more recent retrospective study<sup>12</sup> derived a true incidence of 1.7% of OIS postendovascular thrombectomy and concluded that absent choroid blush is a very sensitive imaging finding (on DSA) that can alert the interventional radiologist to potential development of OIS.

The mechanism of OIS post-mechanical thrombectomy is elusive. The plausible pathophysiology of the same has been attributed to the following mechanisms. One is due to embolization to new territory,<sup>13</sup> where during clot retrieval, it can migrate either proximally or distally to a new unaffected territory (OA) resulting in occlusion of distal OA<sup>3</sup> and compromised orbital vascular supply. However, occlusion of the only OA is not enough to cause all the symptoms of OIS. So, additionally, the presence of an anomalous OA with lack of ECA anastomoses<sup>1</sup> or occlusion of both the OA and anastomoses is essential. Occlusion of distal branches of OA precludes sufficient collateral blood flow. Another mechanism that has been postulated is arterial hypoperfusion of the orbit due to acute occlusion.<sup>1,11</sup>

In our case, it was a routine mechanical thrombectomy procedure without any difficulty encountered in clot

retrieval. Patent OA before and after the procedure was well documented. Left CCA, ECA, and rest of the ICA were also normal. This makes our case enigmatic. We propose that a combination of distal micro thromboembolism and orbital hypoperfusion resulted in OIS in our case.

Incidentally, our patient was COVID-19 positive, whether this has any added influence on the development of OIS is unknown.

MRI signs of OIS include axial proptosis, intraconal and extraconal orbital fat stranding, bulky edematous extraocular muscles, and T2 hyperintensity within of the optic nerve with or without diffusion restriction.<sup>14</sup>

The development of the above-mentioned symptoms in the setting of post-mechanical thrombectomy status entails two differentials—CCF and OIS. Signs of OIS include conjunctival congestion, sensory impairment in V1 distribution of the trigeminal nerve, an audible orbital bruit and venous congestion on fundoscopy, which help to clinically distinguish it from OIS.<sup>3</sup> However, imaging is essential to exclude a CCF. A non-dilated superior ophthalmic vein and a normal cavernous sinus, as seen in our case, excluded the possibility of a CCF.

Most of the symptoms of OIS generally resolve over a reasonably short period of time; however, the loss of vision is permanent, as in our case. Treatment is mainly supportive and varies with the underlying cause.

## Conclusion

The purpose of our article was to highlight the occurrence of OIS, especially post-mechanical thrombectomy, albeit rare that too with a patent OA and to discuss its possible pathophysiological mechanisms. The rarity and severity, especially in the setting of mechanical thrombectomy, warrant treating clinicians and radiologists alike to take cognizance of the entity to ensure adequate preventive measures are in place or to aid in prompt diagnosis and institute timely treatment.

### Ethical Approval

This case report describes a rare diagnosis from routine diagnostic procedures. Hence, approval from the institutional review board was not obtained. Written informed consents for all the procedures were obtained before they were performed. For this type of study, consent for publication is not required as the data to be published is sufficiently anonymized.

### Funding

None.

### Conflict of Interest

None declared.

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