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Amalric choroidal infarction, retinal artery occlusion, and ischemic optic neuropathy: Delayed presentations of traumatic internal carotid artery dissection

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ABSTRACT

Purpose: This case report describes the delayed, uncommon ophthalmic presentations of monocular choroidal ischemia (Amalric triangular sign), ischemic optic neuropathy, central retinal artery occlusion (CRAO), and extraocular motility restriction caused by traumatic internal carotid artery dissection (ICAD) in a young individual.

Observations: A 29-year-old man presented with sudden vision loss in his left eye which had started 7 h earlier. His medical history included a motorcycle accident six months prior, where he struck his chin on the ground and lost consciousness. At that time, he had completely recovered with no complications. On the day the patient reported with vision problem, an ophthalmic examination of the affected eye revealed visual acuity of no perception of light (NPL), restriction of extraocular movement, and relative afferent pupillary defect. Fundus examination showed slightly pale optic disc swelling, macular whitening with a cherry red spot appearance indicating the presence of CRAO, and several whitish triangular patches in the peripheral retina. Fundus fluorescein angiography revealed delayed arm to choroidal and retinal circulations in the early phase, with hyperfluorescence and hyperfluorescent staining along the areas of whitening triangular patches in the later phase. Carotid doppler ultrasonography and magnetic resonance angiography confirmed an extracranial left ICAD. After the 3-month follow up, the patient's vision remained NPL with hypo/hyperpigmentation changes along the previous whitish patches in the peripheral retina.

Conclusion and importance: This case underscores the delayed onset of ocular ischemic symptoms associated with ICAD following head and neck trauma in young individuals. Despite the low risk, patients may need to be informed about the possibility of these late occurring ophthalmic complications and physicians need to stay vigilant for these conditions, which may arise months after the initial trauma.

1. Introduction

Traumatic internal carotid artery dissection (ICAD) is an uncommon condition with heightened susceptibility in individuals who have previously sustained head, facial, or cervical spine injuries.[1,2] Younger individuals are more commonly affected by traumatic ICAD compared to cases with a spontaneous origin. This condition has been recognized as a potential risk factor for stroke in young individuals.[3] However, traumatic ICAD may manifest with diverse clinical features. Some cases remain asymptomatic, while others develop cerebral or ocular ischemic stroke due to hypoperfusion from an intramural hematoma or arterial embolization.[4] Additionally, the majority of traumatic ICAD patients experience neuro-ophthalmic symptoms within hours to days after the initial trauma with a small percentage developing delayed symptoms months or years later.[5]

The classic triad for diagnosing ICAD includes ipsilateral head and neck pain, Horner syndrome, and cerebral or retinal ischemic stroke; nevertheless, not all patients exhibit all of these symptoms. Importantly, some patients only have ocular symptoms as the presenting sign of dissection-induced ischemia, which can lead to a delayed or misdiagnosis.[6,7] Transient monocular vision loss and ipsilateral painful Horner syndrome are the most common ocular manifestations, while persistent vision loss caused by ischemia of the optic nerve, retina, choroid, or oculomotor nerve are far less common. Amalric triangular sign is a rare

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consequence of choroidal infarction that indicates hypoperfusion of the posterior ciliary arteries (PCAs) and their branches, potentially resulting in permanent visual loss. This condition manifests as a whitened wedge or triangular shape in the choroid with its base locating at retinal periphery and its apex pointing toward posterior pole. When fluorescein angiography (FFA) is performed, the affected area appears hypofluorescent in the early phase and hyperfluorescent and staining in the late phase.[8]

The purpose of this study is to present an uncommon instance of a patient who had delayed presentation of sudden vision loss, accompanied by retinal and Amalric choroidal infarction, ischemic optic neuropathy, and oculomotor nerve dysfunction as a result of blunt traumatic ICAD.

2. Case report

A healthy 29-year-old male was referred from a primary care center after experiencing a sudden loss of vision in his left eye while sitting in a car. When he arrived, he indicated that the vision impairment had persisted for approximately 7 h. In addition to vision impairment, he also experienced an ipsilateral throbbing headache. He disclosed that he had a motorcycle accident six months earlier, during which he struck his chin on the ground. He had been unconscious for an hour and appeared to recover without any consequences at that time.

On presentation, the ophthalmic examination revealed visual acuity (VA) of 20/20 in the right eye and no light perception (NPL) in the left eye. The right eye appeared normal, but the left eye showed a relative afferent pupillary defect, an intraocular pressure of 10 mmHg, limited ocular movements in all directions except abduction (Fig. 1). Fundus examination revealed multiple well-defined, deep whitish triangular patches of the choroid, an opaque whitening retina at the posterior pole indicating a cherry red spot, and a pale optic disc swelling (Fig. 2A). Optical coherence tomography (OCT) showed diffuse edema of both the outer and inner retinal layers (Fig. 2B). Fundus fluorescein angiography demonstrated a delayed in choroidal and retinal perfusion in the affected eye. Furthermore, there was an early and late hypofluorescence over the retina along the posterior pole corresponding to the retinal ischemic area, and an early hyperfluorescence with a late staining over the triangular whitening patches, corresponding to the choroidal ischemic area (Fig. 3A–D).

A systemic examination found no associated neurological deficits or anhidrosis. A cerebral computed tomography scan performed at the primary care center showed no signs of cerebral infarction or hemorrhage. Comprehensive laboratory tests, including complete blood count, blood sugar, lipid profile, antinuclear antibody, antiphospholipid antibody, erythrocyte sedimentation rate, C-reactive protein, protein C, and protein S, yielded normal results. Additionally, the electrocardiogram and echocardiogram were normal.

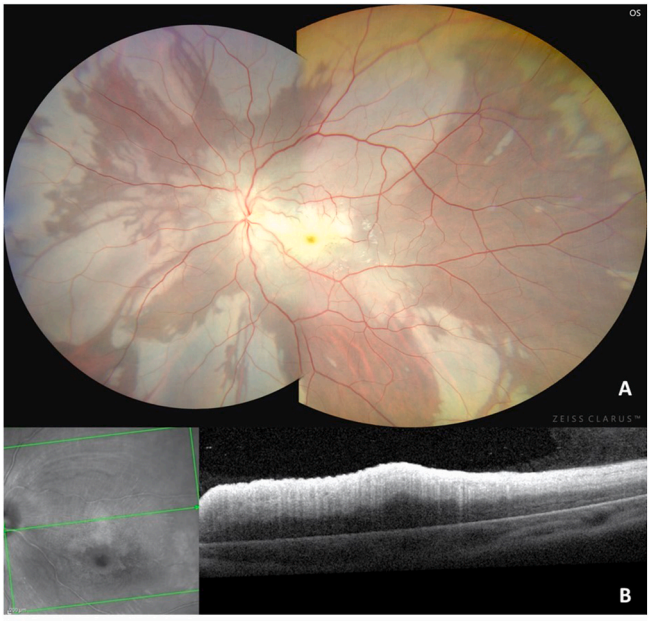

Fig. 2. On presentation, a color fundus photograph illustrating multiple whitish triangular patches in the peripheral retina corresponding to choroidal infarction and retinal whitening over posterior pole corresponding to retinal artery occlusion (A). Optical coherence tomography demonstraing diffuse retinal thickening and loss of entire retinal layer boundaries (B). (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

However, a Doppler ultrasound of the carotid artery demonstrated complete blockage of the left internal carotid artery (ICA) (Fig. 4A and B). Magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) scans of the orbit and brain revealed the dissection of the left carotid bulb extending to the left bifurcation, with severe stenosis to occlusion of the cervical ICA up to the ICA supraclinoid segment (Fig. 5A and B). Diffusion-weighted MRI showed restricted diffusion in

Fig. 1. On presentation, ocular motility showing limitation in almost all directions in the left eye.

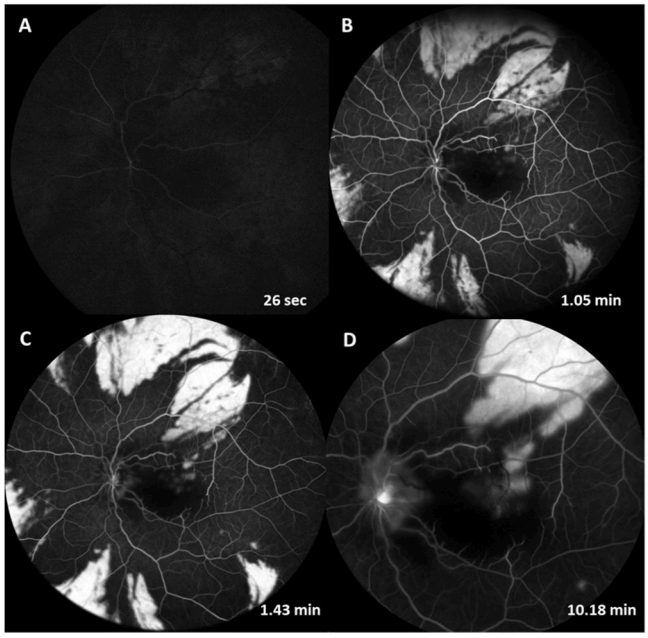

Fig. 3. Fundus fluorescein angiography showing a delayed in choroidal and retinal filling time in an early phase taken at 26 seconds (A) hyperfluorescence of the choroid in the corresponding triangular whitening area on fundus examination and retinal vasuclar filling defect over the macular area taken at 1.05 minutes (B) and 1.43 minutes (C) hyperfluorescent staining of the choroidal infarction area and disc leakage in late phase taken at 10.18 minutes (D).

the left optic nerve and retina, consistent with ischemic optic neuropathy and retinopathy (Fig. 5C). The brain parenchyma appeared normal. Treatment included antiplatelet therapy and high dose statin were given to prevent any subsequent ischemic events.

At the three-month follow-up visit, the patient's vision remained NPL. The fundus examination revealed optic disc atrophy, generalized retinal atrophy, and pigmentary alterations in the retina along the area of the initial triangular choroidal whitening. OCT showed thin, atrophic, and disorganized retinal layers (Fig. 6).

3. Discussion

Traumatic ICAD is an important condition to recognize in young individuals who develop stroke. The ICA gives rise to the ophthalmic artery, which is the main blood supply for the eyeball, extraocular muscles, adnexal tissue, and orbital structures. Consequently, when the ICA is compromised, in addition to cerebral symptoms, various signs of ocular ischemia may manifest.[9–11] This case demonstrated an unusual abrupt onset and persistent visual loss caused by ICAD in a young man who had sustained a head injury in a motorcycle accident six months earlier. The visual symptom was accompanied by an ipsilateral headache. Comprehensive ophthalmic examination revealed several signs of ocular ischemia, including triangular choroidal infarction (Amalric sign), ischemic optic neuropathy, retinal artery occlusion, and restricted ocular motility.

Prior studies reported that the time interval between blunt cervical trauma and the onset of ischemic symptoms related to ischemic ICAD varied. While approximately half of the patients experienced clinical signs and symptoms within the first 24 hours, around one-third developed symptoms months or years later after an asymptomatic period, as shown in this particular study.[12–15] The delayed onset of ischemia might be attributed to thromboemboli detaching from the dissected vascular area. However, in this instance, it was hypothesized that a gradual buildup of intramural and/or subintimal hematoma, even with partial vascular healing, was more likely to contribute to progressive stenosis and ultimate occlusion of the arterial lumen.

Several studies described the ocular manifestations of ICAD. According to Biousse et al., 52 % of ICAD patients, including traumatic and spontaneous in origin, initially presented with neuro-ophthalmic symptoms. Among these individuals, 23 % suffered a stroke within the first week, and 31 % within the first two weeks. Nonetheless, the most prevalent ophthalmic presentations in their study were painful Horner syndrome and transient monocular visual loss, with no patients experiencing permanent visual loss.[16]

Song et al. categorized ophthalmic signs and symptoms associated with ICAD into three groups: visual disturbances, oculosympathetic palsy, and ocular motor palsy, which may occur independently or in combination.[17] The severity of visual disturbances in ICAD patients, ranging from transient to permanent visual impairment, varies depending on the degree and location of vascular branch involvement such as PCAs, central retinal artery (CRA), and muscular branches. Hayreh's study on PCAs reported that ischemia along the distribution of short PCAs, long PCAs, or cilioretinal artery can cause the choroid, macula, retina, and optic nerve head to be particularly susceptible structures.[18]

Amalric triangular choroidal infarction, an uncommon ocular condition, has been reported as one of the causes of vision loss in patients with ICAD.[19,20] Previous studies have shown that unilateral Amalric choroidal infarction can be associated with a number of underlying conditions including in cases of traumatic ICAD, retrobulbar hemorrhage, or cocaine use. Bilateral involvement has also been observed in disseminated intravascular coagulation, intravascular lymphoma, giant cell arteritis, systemic lupus erythematous, Raynaud disease, and polyarteritis nodosa, though not simultaneously.[21–26] The presence of Amalric choroidal infarction indicated reduced blood flow from the ophthalmic artery and/or the distal and long PCAs, which supply the

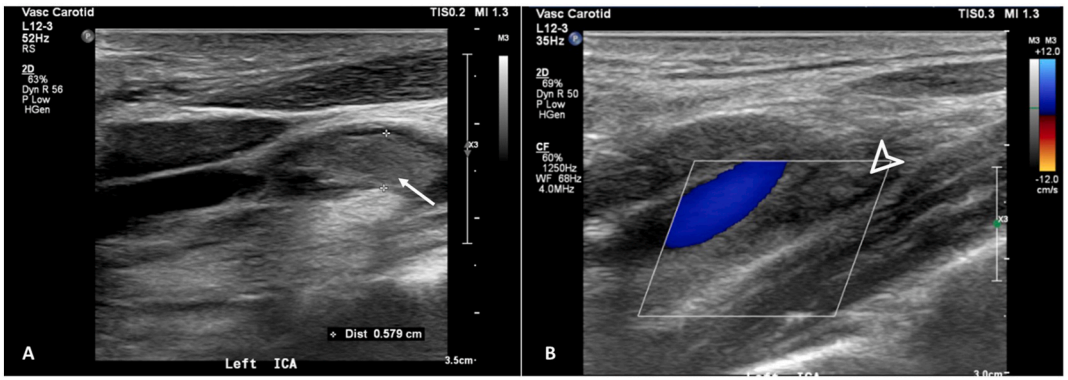

Fig. 4. Carotid doppler ultrasound displaying intraluminal echogenicity within the proximal ICA (arrow) (A) and an absence of flow in the remaining ICA (arrow head) (B).

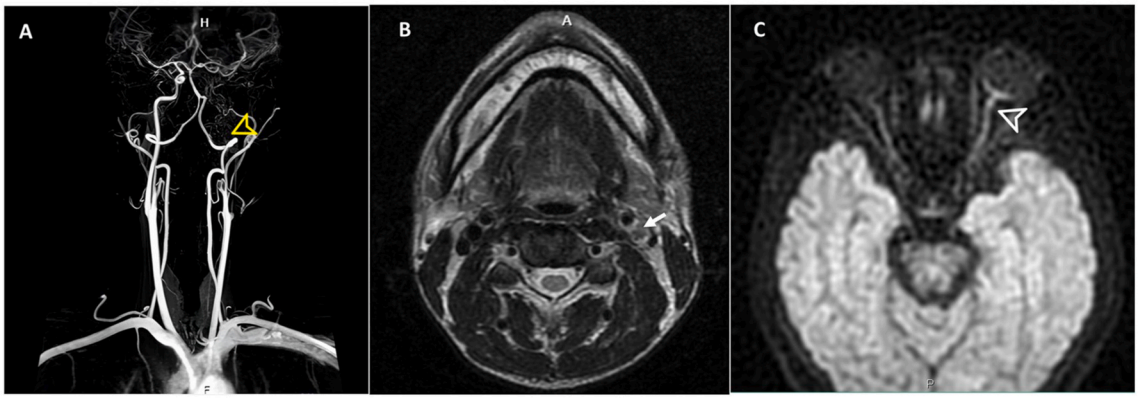

Fig. 5. Magnetic resonance imaging showing an eccentric filling defect along anterior wall of left carotid bulb extending to bifurcation (yellow arrow head) (A) T2-hypointense image showing acute intramural hematoma (arrow) (B) and Diffusion-weighted magnetic resonance imaging (DWI) showing restricted diffusion of the left optic nerve (white arrow head) (C). (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

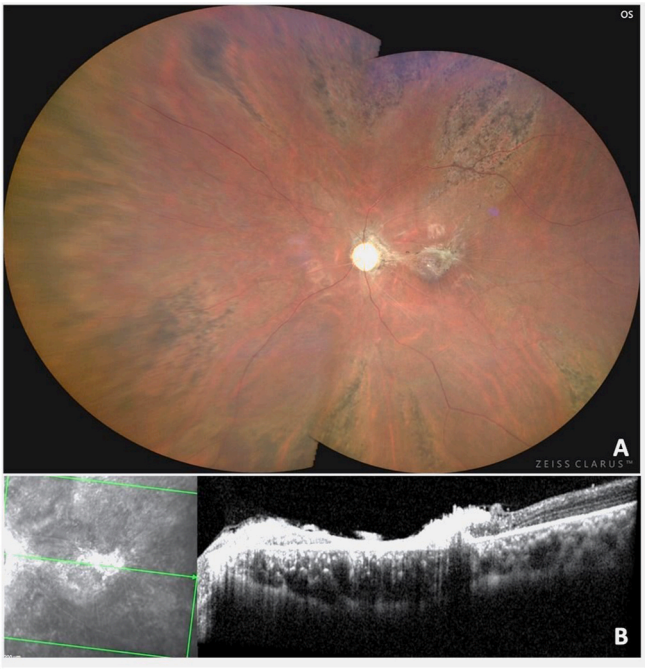

Fig. 6. At three-month follow-up, a color fundus photograph presenting hyperpigmentation in the preceding choroidal ischemic area and optic disc atrophy (A). Optical coherence tomography showing generalized thinning and disorganization of all retinal layers (B). (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

choroid in a wedge-shaped pattern, with the apex directed toward the macular area and the base toward the peripheral fundus. In this particular case, the combination of nonarteritic anterior ischemic optic neuropathy (NA-AION), Amalric choroidal infarction, and restricted extraocular motility may indicate the compromised blood flow in paraoptic PCAs, centrifugal intraneural branches of the CRA, and muscular branches of ophthalmic artery, which is consistent with prior reports.[26–28] Furthermore, the presence of simultaneous CRAO may also suggest an impairment in the CRA circulation.

Nonarteritic posterior ischemic optic neuropathy is another vision-threatening condition associated with ICAD, as described in previous literatures.[7,29,30] However, in the early stages, the affected eye may exhibit visual disturbance, presence of RAPD, and visual field defect indicative of optic neuropathy, despite a normal-appearing fundus and optic disc. Optic disc pallor or atrophy may develop later on. The primary cause is thought to be impairment of tiny collateral arterial branches originating from the ophthalmic artery. Ocular ischemic syndrome (OIS) is another manifestation of ICA stenosis or occlusion that can exhibit several symptoms and signs of compromised blood flow including headache, periocular pain, visual loss, or ophthalmoplegia. However, on fundus examination, OIS reveals attenuation of retinal arteries, scattered dot and blot hemorrhage in the midperipheral retina, and a lack of exudate within macular area. Some patients may develop rubeosis iridis and neovascular glaucoma.[31] Isolated central retinal artery occlusion and isolated oculomotor nerve palsy have also been reported as atypical ophthalmic presentations triggered by an embolic or hemodynamic event following ICAD.[4,30–34]

4. Conclusion

Numerous factors can lead to choroidal and retinal ischemia. Nonetheless, in the event of monocular visual loss accompanied by ocular ischemia affecting various vascular areas in young individuals, post-traumatic ICAD should be warranted as a possible etiology even months or years after the initial event. The patients may need to be advised about the possibility of this late-occurring ophthalmic complication.

CRediT authorship contribution statement

Pavinee Tangkitchot: Writing – review & editing, Writing – original draft, Project administration, Methodology, Data curation, Conceptualization. **Kittisak Unsrisong:** Writing – review & editing, Investigation, Conceptualization. **Janejit Choovuthayakorn:** Writing – review & editing, Supervision, Methodology, Formal analysis, Data curation, Conceptualization.

Patient consent

The patient consent had not been given since the patient had been lost to follow up. This article did not contain any personal information of the patient.

Authorship

All authors attest that they meet the current ICMJE criteria for authorship.

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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