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Case Report

A Case of the Internal Carotid Artery–Posterior Communicating Artery Aneurysm Mimicking Tolosa–Hunt Syndrome

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A 53-year-old woman experienced a right retrobulbar pain followed by ipsilateral extraocular palsies in all directions without dilated pupils or ptosis. Because a plain head computed tomography (CT) scan obtained on her initial visit showed no abnormal findings, such as subarachnoid hemorrhage or a giant cavernous aneurysm, her condition was provisionally diagnosed as Tolosa–Hunt syndrome and elective magnetic resonance (MR) imaging was scheduled. The day after her initial visit, however, she suddenly developed complete ptosis and a dilated pupil on the right side. Emergency MR imaging and angiography revealed a clover leaf-shaped aneurysm projecting to the cavernous sinus at the junction of the internal carotid artery and the posterior communicating artery. Her condition was diagnosed as impending rupture of the aneurysm, and she underwent emergency open surgery. Her symptoms completely resolved within the following 2 weeks. Our case demonstrated that a medium-sized internal carotid artery–posterior communicating artery aneurysm can cause simultaneous oculomotor and abducens nerve palsies with retrobulbar pain if the shape of the aneurysm is complicated. Although these symptoms are very similar to those of Tolosa–Hunt syndrome, we believe that prompt radiological examinations such as MR or 3D CT angiography should be performed to prevent subsequent rupture of the aneurysm.

Keywords: abducens nerve palsy, cerebral aneurysm, oculomotor nerve palsy, retrobulbar pain, Tolosa–Hunt syndrome

Introduction

Ocular pain and oculomotor nerve palsy can occur in various clinical conditions, including orbital lesions, cranial neuropathies, cavernous or parasellar neoplasms, vascular disorders, inflammatory diseases, infectious etiologies, and other illnesses such as cluster headache and migraine.[1–3] In addition, differential diagnoses for painful ophthalmoplegia include compression of cranial nerves by intracranial aneurysms. This is largely regarded as a high-risk condition because acutely developing cranial nerve deficit often precedes rupture of the aneurysm.[4] The most common form of ophthalmoplegia caused by aneurysmal compression is oculomotor nerve palsy.[3] Ophthalmoplegia in all directions due to the mass effect of a medium-sized unruptured aneurysm, and not of a giant intracavernous aneurysm, is extremely rare.[5–7]

In this case report, we describe a patient with a non-giant internal carotid artery–posterior communicating artery (ICA-PCoA) aneurysm presenting with ophthalmoplegia in all directions with concomitant severe ocular pain, thus mimicking Tolosa–Hunt syndrome. The patient's aneurysm had a spindling and irregular shape that presumably enlarged rapidly to cause oculomotor and abducens nerve palsies simultaneously.

Case Report

A 53-year-old woman with no past medical history suddenly started to see some colorful and glaring waves in the upper visual field of the right eye lasting for seconds. A few hours later, she began to feel moderate pain in the back of the right eye. Because she began to have diplopia and photophobia on the fourth day after the onset, she visited our hospital. Although the size of pupils and light reflex were normal and ptosis was not observed, right ocular movement was obviously impaired in all directions. The position of the right eye was slightly deviated laterally, but she could not move it to the right. She had no history of diabetes mellitus. A plain computed tomography (CT) scan of the head reveal no abnormality such as a giant intracavernous aneurysm, subarachnoid hemorrhage, or a parasellar tumor (Fig. 1A). Because her symptoms consisted of painful ophthalmoplegia in all directions, her condition was tentatively diagnosed as Tolosa–Hunt syndrome. We scheduled elective magnetic resonance (MR) imaging for her and sent her to a neurologist at our hospital.

On the next day, however, she developed right eye ptosis and returned to our hospital. At this time, the right pupil was dilated and light reflex was absent on the right side, indicating right complete oculomotor nerve palsy. Her emergency MR images (enhanced T1 3D FLASH, slice thickness 0.8 mm, TR 16 msec TE 4.91 msec) confirmed an irregularly shaped ICA-PCoA aneurysm. A cerebral angiogram showed a vertically long aneurysm measuring approximately 10 mm in its longest dimension with complicated bulges projecting postero-infero-laterally (Fig. 1B, C). No other intracranial abnormalities, including cavernous sinus lesions, were found. On the basis of diagnosis of the ICA-PCoA aneurysm causing oculomotor and abducens nerve palsies, we performed emergency open craniotomy and clipping of the aneurysm. Two-thirds of the aneurysm dome was buried behind the dura and projected toward the lateral cavernous sinus (Fig. 2A). The first bulge of the aneurysm compressed

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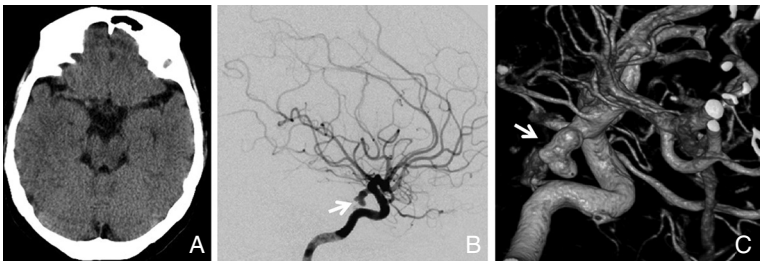

Fig. 1 A: An axial computed tomography (CT) scan of the head showing no subarachnoid hemorrhage. B, C: Right carotid angiograms demonstrating an irregularly shaped aneurysm (arrow) at the junction of the internal carotid artery and the posterior communicating artery.

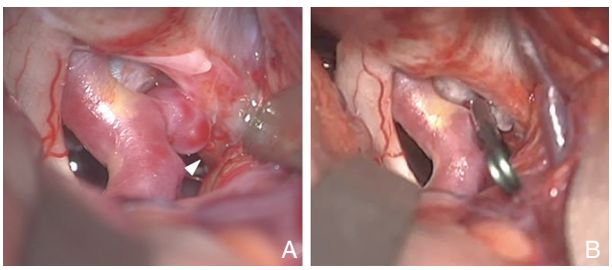

Fig. 2 Intraoperative photographs. A: Only a part of the aneurysm was present in the cisternal portion. The oculomotor nerve (arrowhead) was identified behind the aneurysm. B: The aneurysm was clipped, but the tip of the aneurysm could not be directly confirmed.

the oculomotor nerve. No evidence of subarachnoid hemorrhage was observed, and the aneurysm was successfully obliterated with a clip (Fig. 2B).

The patient showed an uneventful postoperative course with immediate resolution of the retrobulbar pain and dilated pupil within several days. Ophthalmoplegia started to improve 5 days after surgery and showed a complete recovery over the following 2 weeks. On follow-up, her symptoms had disappeared completely, and CT angiography performed 1 year after surgery revealed complete obliteration of the aneurysm.

Discussion

Unruptured cerebral aneurysms can cause cranial nerve palsy due to a mass effect.[8] Among these symptomatic aneurysms, the relationship between ICA-PCoA aneurysms and oculomotor nerve palsy has been well documented in the literature.[5,9,10] Expeditious radiological examinations are warranted for rapidly developing oculomotor nerve palsy because it can be a warning sign of imminent rupture of the aneurysm.[4,8] Retrobulbar pain is frequently associated with this situation and should not be overlooked. Hahn et al. reported that 61% of patients with giant intracavernous aneurysms suffered from retrobulbar pain and speculated that the pain was more frequent because giant aneurysms could cause greater compression or ischemia than smaller ones.[11] However, even small- to medium-sized ICA-PCoA aneurysms can frequently cause ocular pain preceding the onset of oculomotor nerve palsy.[12–17] A previous study reported that 64% of patients with a small ICA-PCoA aneurysm causing oculomotor nerve palsy had antecedent retrobulbar pain.[18] Possible mechanisms of ocular pain associated with non-giant unruptured ICA-PCoA aneurysms include mechanical stimulation to the tentorial incisura due to the adhesion of the aneurysm wall,[12,17] vascular pain owing to rapid aneurysmal growth,[15] and compression of the trigeminal nerve.[1,13,16] Of note, cadaveric and animal studies reported by Lanzino et al. demonstrated that the third cranial nerve is not only an efferent nerve but also contains sensory ganglion cells and conveys afferent fibers.[17] They observed the fibers entering the brain stem and reaching the spinal trigeminal nucleus, indicating that retrobulbar pain might be a form of vascular compression syndrome.

In our case, we first suspected Tolosa–Hunt syndrome because the patient mainly complained of ocular pain and ophthalmoplegia in all directions without pupillary dysfunction. Saito et al. reported three cases of pupil sparing oculomotor nerve paresis caused by unruptured ICA-PCoA aneurysm.[19] They speculated that the narrow and long aneurysm body might result in the confined compression on the oculomotor nerve at a small area where the parasympathetic fibers are scarce. Although it is difficult to determine why the pupillary function was initially preserved in our case, the shape of the aneurysm in our case was compatible with their speculation.

Despite some sporadic case reports on giant or dissecting aneurysms mimicking Tolosa–Hunt syndrome,[20–22] our extensive literature search found only one case of a small ICA-PCoA aneurysm (5 mm × 7 mm) presenting with painful ophthalmoplegia in all directions.[7] The shape of the aneurysm in this report is quite similar to that in our case, namely, the aneurysm was extremely long with complicated bulges projecting infero-postero-laterally. Coronal MR images obtained before surgery demonstrated one of three aneurysmal prominences appeared to compress the right third nerve and another was adjacent to the abducens nerve (Fig. 3). We presume that a non-giant but long aneurysm with this type of projection might cause both oculomotor and abducens nerve palsies. However, it is difficult to determine the exact mechanism by which this would occur—whether it was direct aneurysmal compression to the intracavernous portion of the abducens nerve or a regional increase in the pressure from a minor leak. However, in our case, the postoperative immediate resolution of all symptoms, including both oculomotor and abducens nerve palsies, suggests that the acutely developing symptoms were attributable to the aneurysm.

Our case highlighted that painful ophthalmoplegia in all directions mimicking Tolosa–Hunt syndrome can occur

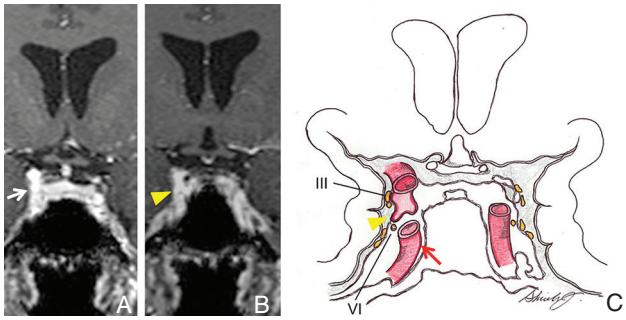

Fig. 3 A, B: Preoperative coronal magnetic resonance (MR) images demonstrating the complicated shape of the aneurysm. Note that one of three aneurysmal prominences (A, white arrow) appeared to compress the right third nerve and another (B, yellow arrowhead) was adjacent to the abducens nerve. C: A schematic drawing showing the aneurysms (yellow arrowhead) and the presumptive location of the cranial nerves. III: oculomotor nerve, VI: abducens nerve, red arrow: the internal carotid artery.

because of a non-giant ICA-PCoA aneurysm with a complicated shape. In other words, the coexistence of abducens nerve palsy does not exclude the possibility of impending rupture of the aneurysm. Prompt radiological examination should be considered because this potentially life-threatening condition, although rare, can be surgically treated if diagnosed properly.

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Conflicts of Interest Disclosure

The authors have no disclosure to report.

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