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CASE REPORT

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Persistent Trigeminal Artery Causing an Abducens Nerve Palsy: A Case Report

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ABSTRACT

We present a case of a 50-year-old female who was diagnosed with an isolated right abducens nerve palsy and was found to have a persistent trigeminal artery (PTA). The trigeminal artery is the most common persistent embryological carotid-vertebrobasilar anastomosis. A PTA can be picked up as an incidental finding on magnetic resonance imaging (MRI) or angiography. It has been reported that a PTA can be found in 0.1 to 0.6% of all cerebral angiograms. PTA has been linked to several rare abnormalities such as vascular aneurysms and cranial nerve compression. Our patient presented with diplopia and was found to have a paresis of the right lateral rectus muscle consistent with a right abducens nerve palsy. MRI found a right-sided PTA indenting the ventral surface of the pons. This case investigates and highlights that neurovascular compression from a PTA can cause an isolated abducens nerve palsy. Further research is required to investigate if surgical intervention for non-aneurysmal PTA would be beneficial for patients.

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Introduction

The trigeminal artery is the most common persistent embryological carotid-vertebrobasilar anastomosis.[1] During development, the trigeminal artery forms when the embryo is approximately 6 weeks old.[2] At this stage, it is the main blood vessel that provides the basilar artery with blood before the development of the vertebral and posterior communicating arteries.[2] The trigeminal artery is usually patent for a period of 7 to 10 days during embryonic development, and after this time it regresses.[3] If the trigeminal artery is present beyond this point, it is classified as a persistent trigeminal artery (PTA) and will remain into adulthood.[3]

A PTA can be picked up as an incidental finding on magnetic resonance imaging (MRI) or angiography. It has been reported that a PTA can be found in 0.1 to 0.6% of all cerebral angiograms.[2,4] A PTA has been linked to several rare abnormalities such as vascular aneurysms and trigeminal neuralgia due to nerve compression.[3] In this report, we discuss a case of PTA causing an isolated abducens nerve palsy.

Case report

A 50-year-old female presented to her local hospital with double vision, that was mainly on right-gaze but intermittently occurred in primary position. She had a history of hypothyroidism, migraine, and an overactive bladder. Her migraines were predominantly right-sided and began at approximately 11 years of age. Her regular medications included levothyroxine, sumatriptan, fluticasone, cetirizine and solifenacin.

She had experienced chronic sinusitis for 5 years, which had been treated with several endoscopic sinus surgery procedures. There was also a history of right-sided hearing loss and tinnitus for 4 years. One year prior to presentation she underwent left-sided sphenopalatine artery ligation for epistaxis.

She was assessed and diagnosed with a right abducens nerve palsy. A non-contrast computed tomography scan of her head was carried out, which identified no abnormalities. MRI was not requested at the time of presentation, as the compatibility of the clip used for the sphenopalatine artery ligation was not known. Her symptoms were managed with a 'Fresnel' prism. She was been given exercises to try to help her control the esophoria, which did not help,

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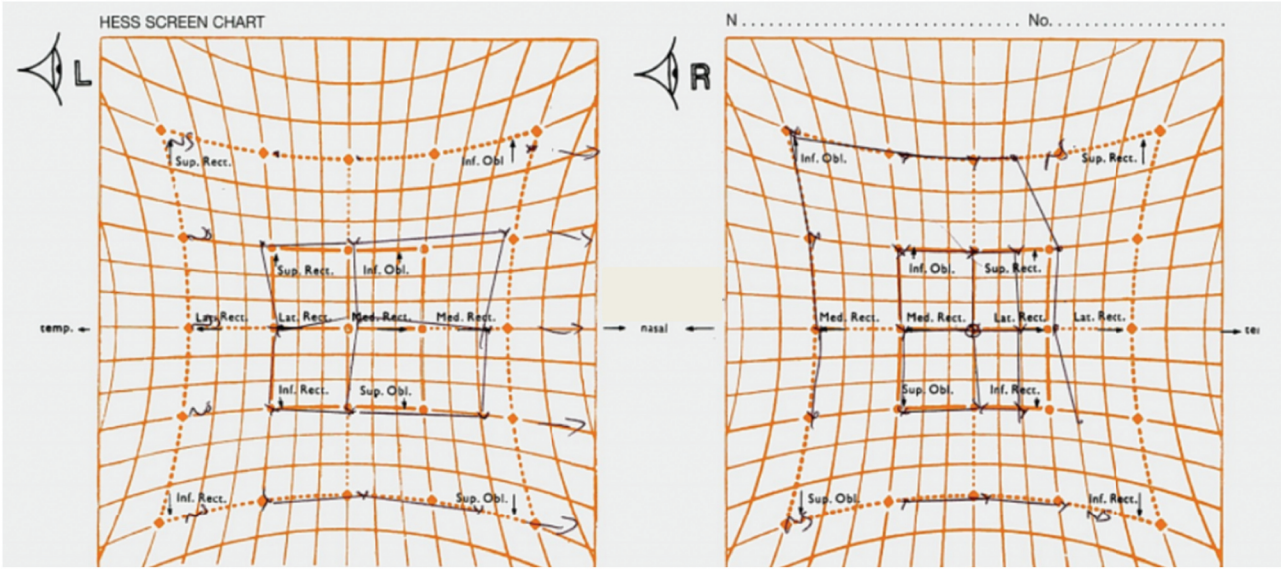

Figure 1. Hess chart showing restricted right eye lateral rectus motility and left eye medial rectus over-action.

so they were discontinued. Due to the coronavirus pandemic, she then experienced delays to her follow-up appointments. At her next appointment, almost 2 years later, there had been some improvement in the abducens nerve palsy. She now had no diplopia in primary position. She was subsequently referred to the neuro-ophthalmology clinic at Lancashire Teaching Hospital NHS Trust (LTHTR) for further evaluation.

On arrival at our department, she reported no change in her symptoms. She wore a 6 dioptre base-in Fresnel prism on her spectacles for driving. Her logMAR visual acuity was 0.18 in her right eye, with no improvement with pinhole, and 0.16 in her left eye, improving to 0.10 with pinhole. A cover test showed a slight esophoria with good recovery (−2 prism dioptres for near and −4 prism dioptres for distance). On ocular motility testing, there was a significant −2.5 restriction of right abduction. A Hess chart confirmed this restriction (Figure 1). She had good stereopsis with 85 seconds of arc using the Frisby Stereotest. The rest of her eye examination including the fundus was normal. Her systemic examination, including a detailed neurological examination of the other cranial nerves, was normal.

We confirmed that the sphenopalatine artery clip was MRI compatible. On the heavily T2-weighted MRI sequence, a prominent arterial structure communicating between the basilar artery and the right cavernous internal carotid artery was demonstrated, suggestive of a PTA (Figure 2). This indented the ventral surface of the pons at the exit location of abducens nerve and was felt to be causing compression of the abducens nerve at that point. A partial empty sella and a few tiny, non-specific foci of T2 hyper-intensities involving white matter was noted, which were felt to be of no significance. She subsequently received extraocular muscle botulinum toxin injections.

Discussion

We found only two other published articles that documented an isolated abducens nerve palsy caused by a non-aneurysmal PTA. Nakamagoe et al. reported a 74-year-old female patient with intermittent diplopia who was found to have an isolated right abducens nerve palsy.[5] They identified that the abducens nerve was sandwiched between the internal carotid artery and a PTA variant. She did not receive any intervention.[5] Kalidindi et al. discussed the case of a 54-year-old female who presented with several months of intermittent diplopia. She was found to have a left-sided PTA compressing the abducens nerve. The PTA joined the cavernous portion of the left internal carotid artery and the distal basilar artery. It was

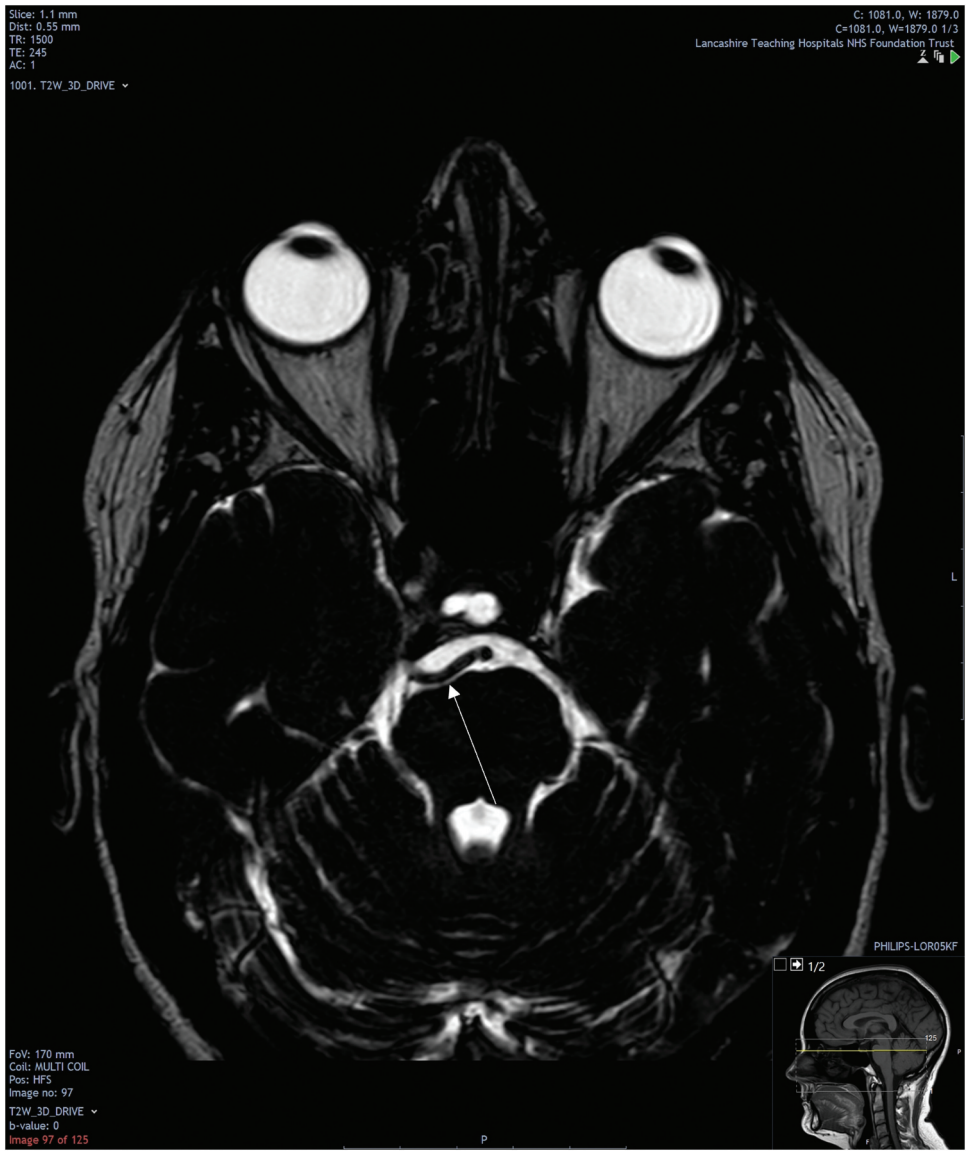

Figure 2. Heavily T2-weighted axial magnetic resonance image showing a persistent trigeminal artery on the right side of prepontine cistern indenting the ventral surface of the right side of the pons (arrow) at the site of the exit of the right abducens nerve.

therefore believed that the compression was occurring in the cavernous sinus. The authors did not comment on the follow up of their patient and did not say if any surgical or medical interventions were conducted.[6] In our case, it is thought that the PTA was compressing the abducens nerve on the ventral surface of the pons at the exit location of the nerve.

It has been documented that PTA is 1.8 times more common in females than in males.[7, 8] The cause of the increased incidence in females is unknown.[7]

Other reported symptoms that can be caused by PTA are trigeminal neuralgia, headaches and cerebral ischaemia.[4,9,10] Our patient had experienced right-sided migraines since a young. The exact pathophysiology of migraine is still unknown. However, there is evidence that the dorsal pons is selectively activated during an episodic migraine attack.[10] It made us consider whether there could be a link between the PTA indenting the ventral surface of the pons and the longstanding episodic migraine attacks in this case. Several research studies investigating patients with episodic migraine used glyceryl trinitrate to trigger migraine attacks and found that the dorsal pons was subsequently activated on MRI.[10–12] This led to the hypothesis that the pons may be the centre of migraine stimulation. More recently however, research has focused

on the role of the hypothalamus in migraine development. Functional MRI studies have shown increased activity between the hypothalamus, pons and spinal trigeminal nucleus suggesting that the hypothalamus network might be the real centre of migraine stimulation.[10,12,13] PTA as a cause of the migraine in this case cannot be excluded. A case report by Li et al. found that a large aneurysmal PTA caused symptoms of a migraine, which later resolved after coil embolisation.[14] Another study by Uhlig et al. documented that there is currently little evidence linking PTA to migraine-type headaches.[15] It is therefore worth noting that the PTA in our case could contribute to the history of longstanding right-sided migraines, but at this stage the research evidence is not strong enough to confirm or reject a significant link.

One of the more commonly documented effects of PTA is trigeminal neuralgia. The reported link between PTA and trigeminal neuralgia is based on its anatomical location. The PTA was given its name due to its proximity to the Gasserian ganglion and trigeminal nerve.[16] If the trigeminal artery fails to regress, it can cause compression of the trigeminal nerve leading to neuralgia symptoms.[16] Kempe and Smith found that a PTA could displace and compress the trigeminal sensory nerve root.[17] De Bondt et al. examined MRI studies from 136 patients presenting with trigeminal neuralgia and found that a PTA was present in 2.2%.[18] The reported rate of an incidental finding of PTA is 0.1–0.6% on cerebral angiography.[4] The prevalence rate found in the de Bondt et al. study of 2.2% is therefore clinically significant, and they concluded by recommending a cerebral MRI or angiogram for patients presenting with trigeminal neuralgia as a non-invasive way to aid the diagnosis.[18]

Although not identified in this case, a PTA can lead to cerebral ischaemia. This was documented in a case report by Palmer and Gulcer in 1981, whereby a PTA was deemed the likely cause for emboli to pass from an ulcerated carotid bifurcation to the vertebro-basilar circulation.[19] Since then, there has been a growing body of evidence linking a PTA with cerebral ischaemia.[4,20–22] It is thought that the PTA allows a channel of communication allowing a plaque from atherosclerotic internal carotid emboli to flow into the vertebrobasilar network. Kwon et al. presented a case of right-sided weakness secondary to cerebral infarction. The patient received thrombolysis with intravenous tissue plasminogen activator. On MRI, it was shown that the patient had a PTA occlusion leading to the cerebral ischaemia. Post-thrombolysis the PTA occlusion resolved and circulation was restored to the basilar artery.[21] This research highlights the importance of the consideration of ischaemic events due to a PTA.

Our patient's symptoms of diplopia were intermittent in primary position, and this has been reported in previous research. One theory is that a transient increase in blood flow through the PTA can lead to temporary nerve compression, which can recover within several weeks.[6,23] Another theory suggests that nerve compression leads to demyelination and once the nerve conduction returns to normal following 'remyelination', the patient's symptoms improve.[5,24] Although the exact mechanism is unknown, these theories are supported by the temporary nature of the diplopia in primary position in our case.

Interestingly, most other research studies found that neurovascular compromise occurred when a PTA was associated with an aneurysm. Ladner et al. discussed a case of trigeminal neuralgia that resolved after PTA aneurysm coiling.[9] Murai et al. presented a case of a PTA aneurysm leading to an abducens nerve palsy. They used coil embolisation with a stent-assisted technique; however, there was no improvement in the cranial nerve palsy after this.[25] The research on the success of coiling therefore has conflicting results and, as with any neurosurgical procedure, there is a risk of further damage to the involved cranial nerve. There have been no reports of treatment of a non-aneurysmal PTA, and consequently our patient has not undergone any invasive surgery for her condition.

Conclusion

We have presented a 50-year-old female with an isolated right abducens nerve palsy thought to be caused by a PTA indenting the ventral surface of the pons. We would recommend MRI for all patients with isolated abducens nerve palsy without vascular risk factors and in all patients with vascular risk factors that do not show signs of recovery.

Further research is required in the future to investigate if surgical intervention for non-aneurysmal PTA would be beneficial for patients.

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