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Case Report

Abducens nerve palsy in a congenital anomalous neurovascular development of the sixth cranial nerve and anterior inferior cerebellar artery: A case report ☆

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ABSTRACT

Abducens nerve palsy is a common ocular motor paralysis with a broad set of etiopathogenetic causes. Magnetic resonance imaging is a key diagnostic technique to investigate organic causes of sixth nerve palsy, as it allows a detailed representation of the course of the nerve, particularly in its intracisternal tract. Anatomical variants of the sixth cranial nerve comprise duplications and fenestrations in various traits. Anatomical variants of cerebellar arteries have also been described. We report the case of a patient with abducens nerve palsy presumably related to a neurovascular conflict due to a peculiar anatomical variant, which consists in a cerebellar artery passing through the intracisternal duplication of the abducens nerve.

Introduction

Abducens nerve palsy is the most common ocular motor paralysis in adults and the second-most common in children, having an annual incidence rate of 11.3/100,000 cases [1].

Although the etiopathogenesis is often undetermined, causes are hypertension alone (19%), coexistent hypertension and diabetes (12%), trauma (12%), multiple sclerosis (7%), neoplasm (5%), diabetes alone (4%), cerebrovascular accident (4%), post-neurosurgery (3%), aneurysm (2%), and miscellaneous causes (8%) [1], including leukemia, migraine and pseudotu-

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mor cerebri [2], the latter being prevalent in young obese females [3].

MRI is a crucial examination to investigate organic causes of sixth nerve palsy and can reveal possible anatomical variants of the nerve. Among the latter, nerve duplication is predominant, and its incidence ranges from 5% to 28.6% in different studies [4]. Not only can MRI identify the causes of an acute deficit of the sixth cranial nerve, but it also allows an accurate morphological evaluation of its intracisternal course. In particular, high-resolution steady state free precession sequences, such as Fast Imaging Employing Steady-state Acquisition, take advantage of the high contrast between nervous tissue and cerebrospinal fluid to depict with high accuracy the anatomy of the intracisternal tract of the nerve, despite providing limited morphological information about cavernous and orbital nerve tracts [5].

We present the case of a patient who was admitted to our emergency department with recently onset diplopia, presumably related to a neurovascular conflict due to the passage of a cerebellar artery through an intracisternal duplication of the abducens nerve. To the best of our knowledge, this is the first case report in literature that identifies this anatomical variant of the abducens nerve combined with a cerebellar artery passing through the nerve duplication.

Case report

A 62-year-old woman without prior neurological or ophthalmological medical history showed up at the emergency room having diplopia for 2 days. The patient had no diagnosed conditions, except for hypertension in good control with propranolol therapy. No Horner's syndrome was noted. Consensual and accommodative reflexes were normal. Funduscopic examination with pupil dilation did not reveal papilledema. A defect of the left eye's abduction due to paresis of the external rectus muscle was revealed. The examination of the other cranial nerves and general neurologic exam were normal. The non-contrast CT scan at admission was unremarkable. An MRI examination, performed on the third day after admission, ruled out any organic pathology at the level of the dorsal pons, in the site of the nucleus of the left sixth nerve. On the same side, no expansive lesions were found in prepontine cistern, cavernous sinus and orbit. The left lateral rectus muscle showed regular signal and morphology. A Fast Imaging Employing Steady-state Acquisition sequence targeting the posterior cranial fossa showed the duplication of the intracisternal tract of the left abducens nerve and a vascular structure passing through the nerve fenestration.

Time-of-flight and post-contrast fast spoiled gradient echo sequences demonstrated an asymmetric duplication of the left anterior inferior cerebellar artery (AICA), with 2 separate branches emerging from the basilar artery: the first and larger arterial branch ran through the cerebellopontine angle cistern, under the internal auditory canal; the second arterial branch passed through the abducens nerve fenestration and ran through the cerebellopontine angle cistern looping inside the internal auditory canal (Fig. 1).

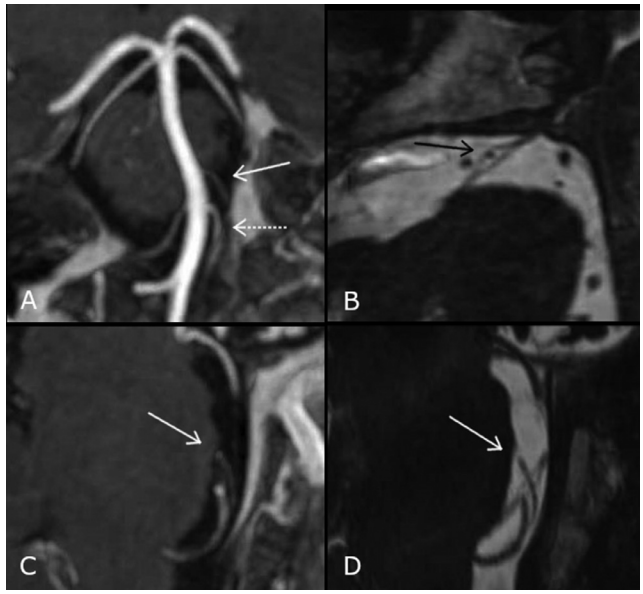

Fig. 1 – Post-contrast FSPGR coronal reformatted image (A) depicting the left AICA duplication, with a major branch emerging caudally (dotted white arrow) and a minor branch emerging cranially (white arrow). An oblique reconstruction of FIESTA image (B) showing the fenestration (black arrow) of the VI cranial nerve in his intracisternal tract, with a vascular structure passing through it. Post-contrast FSPGR (C) and FIESTA (D) oblique reconstructed images demonstrating that the vasal structure corresponds to the minor branch of the AICA (white arrow). AICA, anterior inferior cerebellar artery; FIESTA, Fast Imaging Employing Steady-state Acquisition; FSPGR, fast spoiled gradient echo.

The presence of a neurovascular conflict between the smaller branch of the left AICA and the homolateral sixth cranial nerve at the level of the fenestration was hypothesized, and a possible triggering role of a blood pressure peak was suggested.

The patient was dismissed with a diagnosis of left abducens nerve palsy and a therapy with cardioaspirin and B vitamins was introduced. After 1 month, symptoms were slightly ameliorated but still present.

Discussion

There are several anatomical variants of the sixth cranial nerve, which can be divided into 3 classes [6]: in 70% of cases, the nerve runs as a full-length single trunk from the brainstem to the superior orbital fissure (variant I); in 20% of cases, the nerve passes through the subarachnoid space and in the Dorello's canal as a single trunk, duplicating into 2 branches inside the cavernous space (variant II); in 10% of cases there is a duplication of the intracisternal tract of the nerve (variant III). Wysiadecki et al. [4] described further possible morphological variations of sixth cranial nerve depending on where the duplication occurs: the nerve can emerge from the pon-

tine surface directly with 2 duplicated branches, which can either remain separate throughout their course or join into a single trunk in the cisternal region, in cavernous space or at the level of superior orbital fissure; alternatively, the nerve emerges as a single trunk and then splits into 2 branches at the level of cisternal/petroclival segment or at the level of cisternal/petroclival/cavernous segment.

While the description of morphological variations is usually made on post-mortem specimens, we reported an anatomical variant of the sixth cranial nerve found in vivo on MRI.

In our patient, the splitting of the intracisternal tract of the sixth cranial nerve was associated with the presence of an AICA duplication, whose smaller branch passed through the nerve fenestration.

The combined presence of these anatomical variants could have led to a neurovascular conflict, in which arterial hypertension could have played a role in increasing the chance of developing neurovascular compression, with consequential demyelination and dysfunction of the nerve [7].

The embryology of such a complex anomaly can only be supposed. During the embryogenesis, the forerunner of the basilar artery (the so-called "longitudinal neural system") gives rise to numerous transverse vessels (named "transverse pontine arteries"), each having the potential to become the AICA, depending on the demand of developing cerebellum and brainstem. Peripheric parenchymal demand would be the key to the selection of one or more major branches, leading to the final anatomical configuration of the AICA. This mechanism explains most of the congenital variations of arteries emerging from the basilar artery [8]. Regarding the nerve variant, several authors support the theory that the lateral rectus muscle may be composed of 2 functionally distinct compartments, superior and inferior, which could be independently controlled and selectively activated by the nervous system, supporting a possible nerve duplication [4]. The assumption is that the 2 muscular compartments could drive the development of 2 precursor branches of the nerve that would merge with each other during embryogenesis. Hence, duplication of the abducens nerve might result from alternative developmental pathways.

In conclusion, it is possible to hypothesize that a primitive transverse pontine artery, which subsequently became the smaller branch of the definitive AICA, was enclosed between the 2 different precursor branches of the ipsilateral sixth cranial nerve during their fusion.

Concerning our case, no certain causes of abducens nerve palsy were identified. However, it could be hypothesized that this previously unreported neurovascular variant could have promoted a neurovascular conflict, that may have been exacerbated by a blood pressure peak in a patient with long standing hypertension.

Patient consent

The patient gave the written informed consent for publication.

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