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Case Report

A rare case of ispilateral hemiparesis in a patient with uncrossed pyramidal tract shown by tractography ☆

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ABSTRACT

This article presents a unique case of ipsilateral hemiparesis in a 66-year-old individual, contrary to the conventional understanding of supratentorial strokes causing contralateral neurological deficits. The patient exhibited persistent weakness and sensory abnormalities on the left side of the body following a left occipital infarct. Neuroimaging revealed a chronic stroke in the left occipital lobe, with diffusion tensor imaging demonstrating uncrossed pyramidal tracts at the level of the medulla. The discussion encompasses the anatomical basis of corticospinal tract crossing, historical perspectives, and previous documented cases of ipsilateral strokes. The rarity of complete uncrossed corticospinal tracts without underlying congenital abnormalities or genetic disorders is highlighted. The study underscores the importance of considering such atypical presentations in stroke evaluations and the role of advanced imaging techniques in confirming diagnosis and understanding underlying mechanisms.

Introduction

A supratentorial stroke is widely recognized for causing neurological impairment that affects movement on the contralateral side of the body. This occurrence is attributed to the prevalence of contralateral corticospinal projections originating from the cortical regions of the brain. These projections typically cross in the caudal medulla before descending to the spinal cord. However, contrary to this widely accepted understanding, some studies propose an alternative perspective. There have been a few instances documented in the literature describing strokes with ipsilateral motor deficits.

In this report, we present the case of a 66-year-old individual who exhibited ipsilateral hemiparesis. This unusual manifestation was attributed to an uncrossed corticospinal tract

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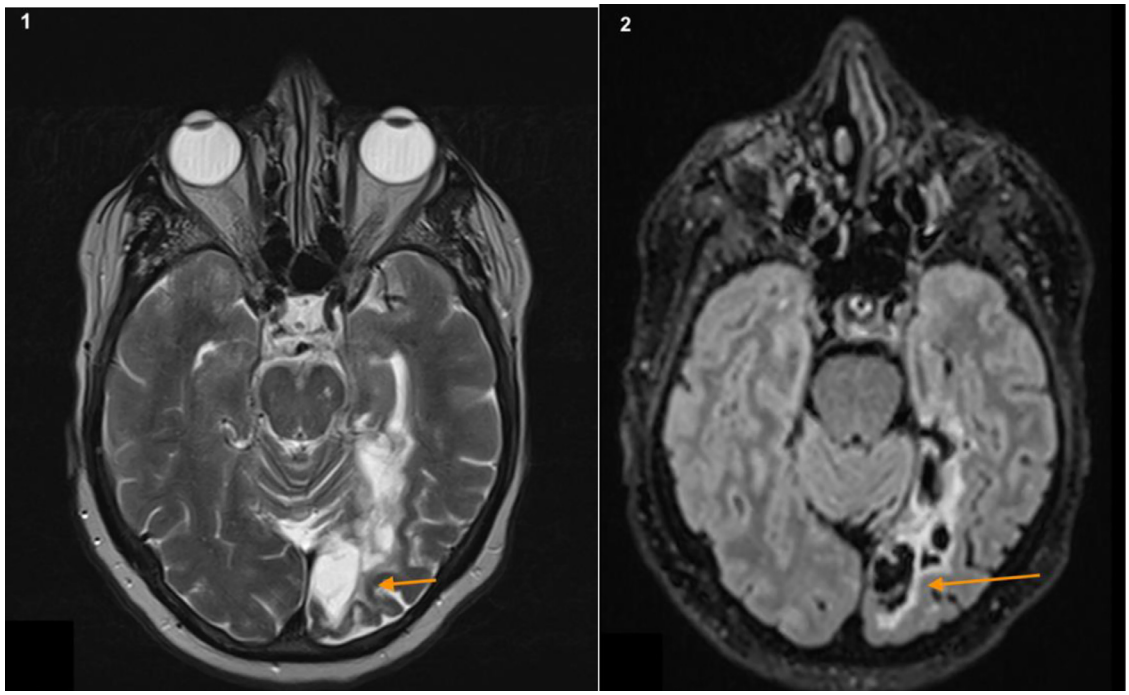

Figs. 1 and 2 – Axial T2-WI and FLAIR showing a well-defined left occipital area hyperintense on T2-WI (Fig. 1) and hypointense on FLAIR (Fig. 2), surrounded by gliosis in hyperintense signal on FLAIR images. Those images are compatible with a chronic ischemic infarct on the territory of the left posterior cerebral artery.

(CST), as revealed by tractography. We have incorporated a brief literature review to enhance comprehension of this phenomenon.

Case report

We report the case of a 66-year-old man, who had history of left hemiparesis and hemianopsia 10 months prior due to a left occipital infarct. He was referred to our department for further evaluation of residual hemiparesis. Despite partial recovery, there were residual neurological deficits. Indeed, the patient exhibited persistent weakness in the left arm and leg, impaired fine motor skills, and numbness on the left side of the face and body. Visual function was compromised, with the left eye showing decreased acuity and ongoing left homonymous hemianopsia affecting the left visual field. Coordination and balance improvements were noted, though mild ataxia persisted in the right arm. Reflexes were brisk on the left side, and cognitive functions remained generally intact, with occasional word-finding difficulty. Speech was fluent but involved mild articulation challenges on the right side. The patient was functionally independent for basic activities of daily living but still required some assistance. An MRI study was performed, and showed in the left occipital a well-defined area hyperintense on T2 WI (Fig. 1) and hypointense on T1-WI and FLAIR (Fig. 2), surrounded by gliosis in hyperintense signal on FLAIR images. The TOF MRA demonstrated flow voids within the left posterior cerebral artery (Fig. 3). All of those findings are consistent with a chronic stroke. The DWI was unremarkable, so

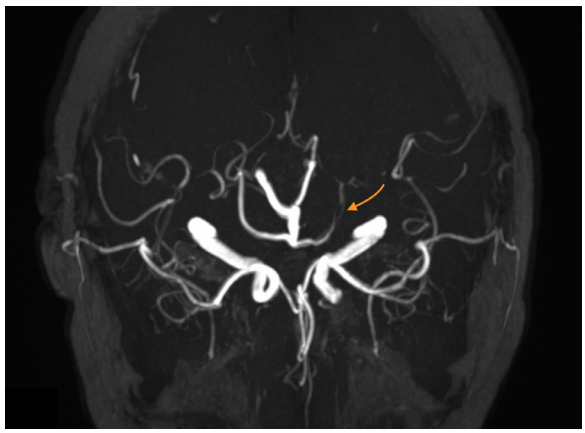

Fig. 3 – TOF MRA demonstrating flow interruption within the second portion of left posterior cerebral artery.

there was no acute infarct. To comprehend the origin of ipsilateral stroke, we proceeded with diffusion tensor imaging. Fiber tracking (Fig. 4A-E) demonstrated uncrossed pyramidal tracts on either side at the level of medulla.

Discussion

We reported here a rare case of ipsilateral stroke due to crossing pathologies of the cortico-spinal tract. Supratentorial stroke is commonly known for causing contralateral neu-

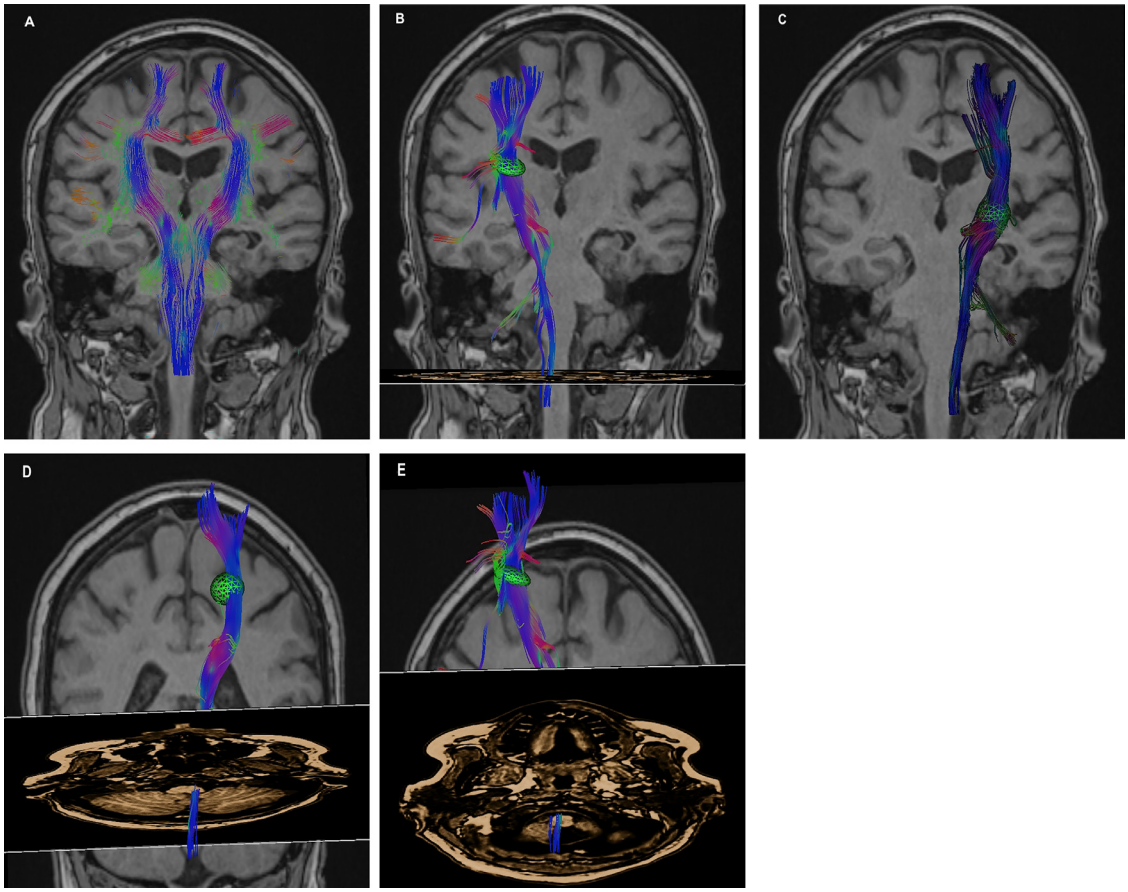

Fig – 4 (A-E) Diffusion tensor imaging (DTI) and 3D tractography showing complete uncrossed pyramidal tracts.

rological deficit. The explanation for that phenomenon being the crossing of the corticospinal tract (CST) at the level of the medulla oblongata.

The axons of the corticospinal projection originate from cells in layer V of multiple regions of the neocortex [1]. The distribution of cortical motor neurons is visually represented by the "motor homunculus," depicting the body arched over the cortical surface to illustrate the relative cortical areas dedicated to different body regions. Approximately half of the corticospinal fibers originate from the primary motor cortex, with the rest arising from various regions, including premotor and somatosensory cortices. In the internal capsule, corticospinal tract fibers maintain a somatotopic order, reflecting a precise arrangement correlated with the body parts they serve. The corticospinal tract makes its entry into the brainstem via the cerebral peduncles and traverses the pons, navigating between the transverse pontine fibers. As it reaches the medulla oblongata, the bundle of corticospinal fibers forms an anterior ridge known as the pyramid. Just before entering the spinal cord, a significant portion of these fibers undergoes decussation, crossing from one side to the other at the pyramids. Subsequently, they enter the lateral column of the spinal cord. A portion of the fibers do not decussate, the majority of which form the anterior column of the spinal cord. However, a smaller proportion of uncrossed fibers contribute to the lateral column on the same side [2].

The crossing of the pyramids was described for the first time by Mistichelli (1709) and François Pourfour du Petit (1710), although the law of cruciate conduction had been known since hippocrate. But it wasn't until Flechsig (1876) came up with his "myelogenetic method" that neurohistological evidence for corticospinal crossing and tract asymmetry emerged. He described variations in the degree and patterns of the results of the crossing. Rakovlev and Rakic confirmed Flechsig results on a larger cohort: Most patients exhibited the bilateral partial decussation; the most common deviation from the standard was bilateral complete decussation; some unusual patterns (Fig. 5) were observed in the brains with congenital abnormalities [3].

Abnormalities of the pyramidal tract are thought to manifest at different developmental stages and typically accompany broader malformations of the brain. They may arise from induction defects, abnormal cell proliferation, irregular neuronal migration, disruptions in axon guidance mechanisms, injury-induced destructive lesions, and malformations leading to abnormalities in decussation [4].

Many studies have reported uncrossed CST associated with other malformations or genetic mutations. Ng et al. introduced a patient diagnosed with Horizontal gaze palsy and progressive scoliosis (HGPPS) who encountered a pure motor stroke on the left side due to an infarction in the left corona radiata. Diffusion tensor imaging tractography established the

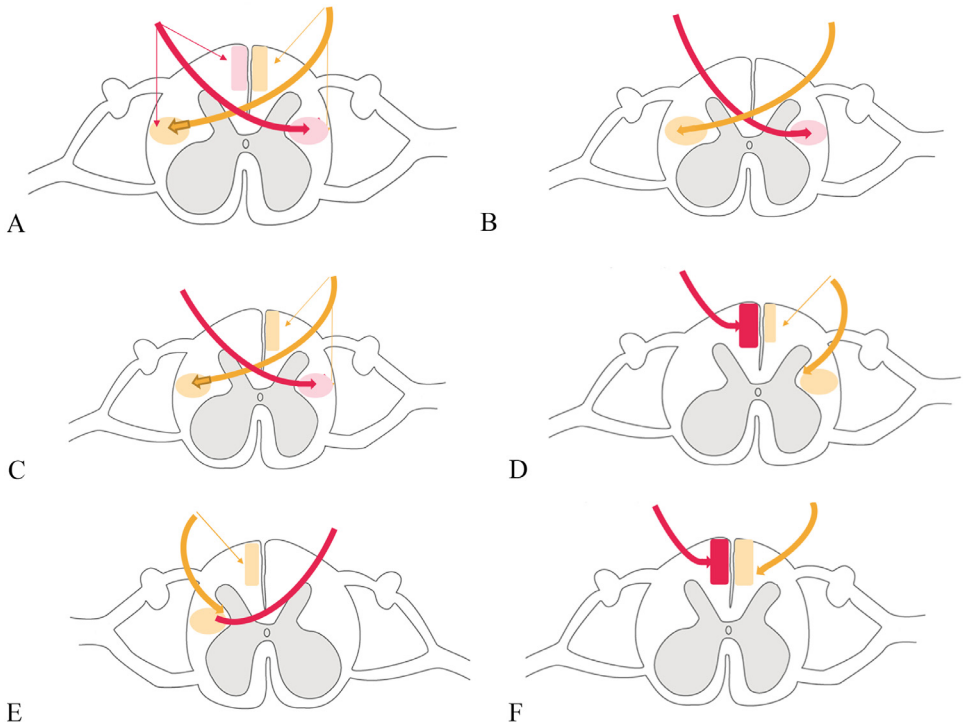

Fig. 5 – The potential variations in the decussation of the pyramidal tracts were observed. In 66.9% of cases, there was a partial decussation, resulting in a larger crossed and a smaller uncrossed pyramidal tract on both sides (A). A complete decussation of both pyramids with the absence of both anterior pyramidal tracts occurred in 16.2% (B). In 13.9%, one pyramidal tract exhibited complete crossing (C). Interestingly, a complete decussation of the left pyramidal tract was six times more frequent than that of the right pyramid. Yakovlev and Rakic's series did not report complete non-decussation of one pyramid (D), although other sources have noted this phenomenon [103]. In a specific specimen, the lateral and anterior pyramidal tracts were absent on the side of the completely crossed pyramid (E). Additionally, in three specimens (2.3%), there was a complete absence of decussating bundles, leading to the absence of both lateral pyramidal tracts (F).

existence of uncrossed corticospinal tracts, explaining the ipsilateral deficit. Additionally, genetic testing uncovered a previously unidentified stop codon mutation in the ROBO3 gene [5]. Amoiridis et al. also presented two brothers with that same genetic mutation, in which uncrossed CST was demonstrated on motor evoked potentials [6]. Kang and Choi described the case of a patient who manifested ipsilateral hemiparesis and spontaneous horizontal nystagmus associated with an agenesis of the corpus callosum [7]. Lagger et al. described two cases of absent pyramid decussation associate to a Dandy-Walker syndrome on postmortem examinations [8].

We described a case of complete absence of crossing on both sides, in a previously healthy patient with no congenital brain abnormality or genetic disorder. To our knowledge, only seven similar cases were described in the literature. All seven individuals had an unremarkable medical history with no notable prior neurologic conditions.

Among these cases, five patients exhibited stroke symptoms and ipsilateral hemiparesis. Another case involved a cerebral tumor with ipsilateral paresis, while one patient displayed ipsilateral motor evoked potentials during intraoperative monitoring of vestibular schwannoma surgery.

Cuatico's initial study in 1979 proposed ipsilateral motor innervation based on electroencephalography, and pneumoencephalography.

In 1996, Hosakawa et al. [9] demonstrated an uncrossed corticospinal tract through somatosensory and motor evoked potentials, with subsequent MRI revealing ipsilateral Wallerian degeneration following a 10-month prior internal capsule and thalamic hemorrhage.

A later case by Terakawa et al. [10] in 2000 described ipsilateral hemiparesis after putaminal hemorrhage, with functional MRI disclosing ipsilateral activation during finger tapping.

The sole instances visualizing outlining a completely ipsilateral course of the CST through diffusion tensor imaging and fiber tracking were reported by Alurkar et al. in 2012 [11] in a patient who presented an ipsilateral stroke due to an underlying right internal carotid artery; Ku et al. in 2017 [12] who incidentally discovered during intraoperative monitoring while performing a microsurgical resection of vestibular schwannoma that electrical stimulation of the frontal scalp triggered motor evoked potentials in the ipsilateral arm; and finally Persad et al. in 2021 [13] whose patient suffered from a left frontal tumors that caused an ipsilateral hemiparesis.

Ipsilateral strokes do not exclusively result from uncrossed corticospinal fibers; some may stem from cortical reorganization within the motor regions of the unaffected hemisphere. Individuals with prior infarcts exhibit adaptive compensation for damaged or disconnected areas within the injured region. It is crucial to investigate previous infarctions, underlying con-

genital anomalies, or functional reorganization of the cerebral cortex in such cases. In the future, these factors may significantly influence patients' prospects for recovery [14,15].

Inatomi et al. investigated the different pathomecanisms of ipsilateral motor deficit and divided them into three groups: Type I refers to an injury of the corticospinal tract in individuals who inherently lack decussation of the tract. Type II signifies an injury to the uncrossed (lateral) corticospinal tract in patients possessing both active crossed and uncrossed tracts. Subtype IIa designates an injury specifically to the uncrossed corticospinal tract, occurring in individuals with both active crossed and uncrossed tracts, that emerges secondarily to a previous injury of the crossed tract [14]. Patra et al. revised these pathomecanisms and added two more: the involvement of ipsilateral fibers with double decussation, and the Involvement of supplemental motor areas [15,16].

The different possibilities are summarized in Fig. 5. Our case was a patient who presented complete uncrossed cortico-spinal tract with no other congenital malformation or abnormality.

Conclusion

This case report details a rare instance of an ipsilateral stroke featuring uncrossed pyramidal tracts, on a previously healthy patient. Clinical confirmation of uncrossed pyramidal tracts was evident in the patient's presentation with an ipsilateral stroke on the same side as the hypodensity on the CT. The use of tractography assisted in documenting the presence of uncrossed pyramidal fibers, reinforcing the accuracy of the diagnosis.

Patient consent

Written informed consent for the publication of this case report was obtained from the patient.

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