ORIGINAL SCIENTIFIC ARTICLES

Thirteen-and-a-Half Syndrome Stroke: A Case Report

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ABSTRACT

We describe a 52-year old woman who developed one- and-a-half syndrome with an ipsilateral trigeminal and facial nerve palsy from a lacunar infarct of the left paramedian pontine area likely involving the median-paramedian perforators of the basilar artery.

Keyword: Stroke, Posterior circulation stroke, one and a half syndrome, thirteen and a half syndrome, lacunar infarct, brainstem stroke syndrome

BACKGROUND

Brainstem syndromes are named in terms of numerical neuro-ophthalmological manifestations. The amalgamation of cranial nerve deficits would define these syndromes. The first written report regarding a unilateral horizontal gaze palsy associated with an ipsilateral internuclear ophthalmoplegia (INO) was by Miller Fisher in 1967.[1] Collectively, this is referred to as one-and-a-half syndrome.[2] This myriad of symptoms would manifest in a patient with a lesion in the ipsilateral tegmentum of the pons. [3]

This case report discusses a rare variant of a one-and-one-half syndrome with an ipsilateral facial and trigeminal nerve deficit, or thirteen and a half syndrome secondary to a lesion involving a basilar artery perforator, probably the a left median-paramedian perforator. Up to this date, there have only been two reported cases of this syndrome. [4]

CASE PRESENTATION

A 58-year-old female presented to the emergency department (ED) 12 hours post-ictus with horizontal diplopia, facial asymmetry, and numbness of the left face. She had no known comorbidities but at the ED, had a sustained elevated blood pressure of 160/80 mmHg. She also had a BMI of 25.3, overweight by Asian standards. Other physical examination and neurologic findings were essentially normal. On laboratory work-up, she had a high capillary blood sugar level at 364mg/dl and an elevated HbA1C level at 9%. She denies illicit drug use, and is a non-smoker and non-alcoholic beverage drinker.

Neurologic examination reveals a normal mental status examination and higher cortical function tests. Cranial nerve examination showed a left internuclear ophthalmoplegia, ipsilateral facial hemisensory loss and a left peripheral facial palsy. There was a remarkable reduction in the left eye abduction with right-beating nystagmus of the right eye on adduction (Fig. 1). Other cranial nerves were intact; with no motor and sensory deficits.

Cranial magnetic resonance imaging (MRI) showed a linear hyperintense signal within the medial aspect on the tegmentum of the pons on DWI with a corresponding signal

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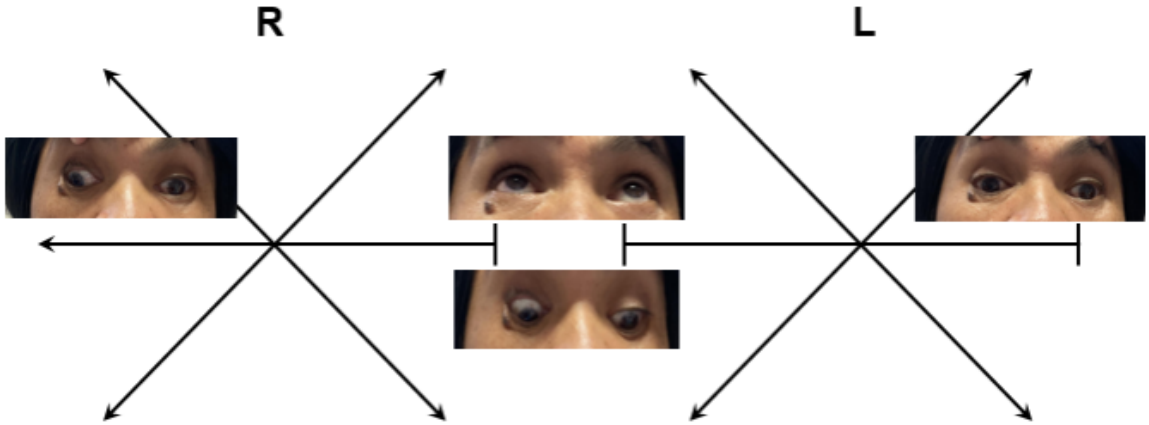
Figure 1. The patient had left gaze palsy and left INO which is seen in a one-and-a-half syndrome.



drop on ADC, involving the medial longitudinal fasciculus, left trigeminal, and left facial nerve (Figure 2). On magnetic resonance angiogram (MRA) done as time of flight (TOF) noted unremarkable results.

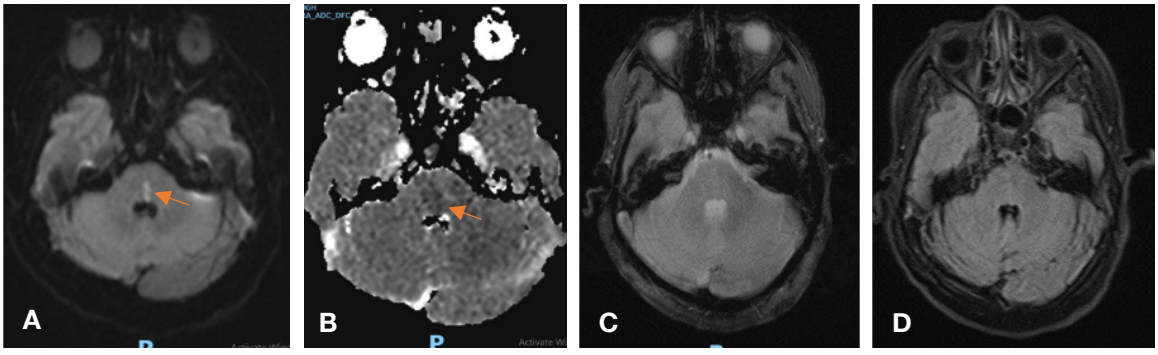
The patient was managed as a pontine infarct and started with double anti-platelet therapy. She was discharged to rehabilitation care after 7 days with stable deficits.

DISCUSSION

Different neurologic deficits of the eye and other cranial nerves can be combined and called according to the involved cranial nerves such as one-and-a-half syndrome, eight-and-a-half syndrome, thirteen-and-a-half syndrome and so on. In this case, the patient presented with a one-and-one-half syndrome with an ipsilateral cranial nerve (CN) V and CN VII palsy, or a thirteen and a half syndrome (7+5+1½=13½) due to an infarct at the parapontine area.

The blood supply of the pons comes from the basilar artery and is divided into 3 groups; median-paramedian and the short and long circumferential.[5] The median-

Figure 2. Cranial Axial Magnetic Resonance: DWI (A) and ADC (B) sequences reveals hyperintensity within the tegmentum of the pons on its medial aspect and a signal drop respectively. GRE (C) and T2 Flair (D) sequences showed unremarkable findings.



paramedian perforators are small branches that arise from the posterior surface of the basilar artery and are not recognizable on angiogram. The majority of these enter the pons at the level of the basilar sulcus and some of these arteries are long enough and traverse the anterior-posterior diameter of the pons to reach the floor of the fourth ventricle. They typically supply the median-paramedian and partially the pons lateral aspect. [6]

Lesions of the paramedian pontine tegmentum, affect the abducens nuclei, medial longitudinal fasciculi and paramedian pontine reticular formation. Being terminal end arteries, they are vulnerable to ischemia in this region. The most common site of pontine lesions involves the basis pontis on one side and often medially in the distribution of the paramedian branches. Infarcts of the pons are the most frequent lesion found in diabetic patients on necropsies and are primarily caused by atheromatous branch disease. [7]

There are only 2 other cases reported of thirteen-and-one-half syndrome (7+5+1½=13½). A case of a post-transplant lymphoproliferative disorder[3] and a case of a severe traumatic brain injury from a motor vehicular accident. [8] In this case, etiology is an acute ischemic stroke involving the paramedian pontine area.

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