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Case Report

Fifteen-and-a-half syndrome: A rare presentation of pontine infarction

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1. Introduction

The one-and-a-half syndrome(OHS) combines a horizontal gaze palsy and internuclear ophthalmoplegia was first described by Miller-Fisher in 1967. It is caused by lesions in the dorsomedial pontine tegmentum, which affect the medial longitudinal fasciculus (MLF) and paramedian pontine reticular formation(PPRF). It is characterized that one eye is unable to move horizontally, while the other one is absent in adduction and abduction is preserved. The rare case of eight -and-a-half syndrome is OHS syndrome plus ipsilateral seventh (facial) cranial nerve palsy which was reported in detail by Eggenberger in 1998 [1].

The fifteen-and-a-half syndrome was first described in 2005 by Bae [2], it is attributed to a tegmental pontine lesions consisting of OHS plus bilateral seventh nerve palsy (1½ + 7 + 7 = 15½). The dorsal tegmentum of the caudal pons involving MLF, PPRF, abducens nucleus and the adjacent facial nerve is the anatomical basis of one-and- a-half syndrome and bilateral peripheral facial paralysis.

Due to its extreme rarity, the clinical course and radiological characteristics of this specific disease are not well-established. In this document, we report one case of a 55-year-old man who presented with fifteen-and-a-half syndrome in which the brain magnetic resonance imaging (MRI) revealed a pontine tegmentum infarction.To our knowledge, this is the first report of fifteen-and-a-half syndrome of an Asian who was admitted to the hospital because of eight-and-a-half syndrome.

2. Case report

A 55-year-old male presented with sudden onset of dizziness, double vision, and slurred speech. He had a history of hypertension and poorly-controlled diabetes mellitus. On admission, his blood pressure was 170/111 mm Hg, the heart rate was 82/minute, regular, and respiratory rate was 24/minute. Neurological examination revealed combination of left gaze paresis and left internuclear ophthalmoplegia. Right eye abduction and vertical eye movements were preserved. Horizontal nystagmus on abduction of the right eye was also observed. The pupils were equal in size and normally reactive to light. Left peripheral facial palsy was also noted (Fig. 1A–F). The brain MRI on hospital day 2 revealed an acute pontine tegmentum infarct which diffusion-weighted imaging (DWI) and fluid attenuated inversion recovery (FLAIR) showed hyperintensity in the dorsomedial pontine tegmentum (Fig. 2A–B).

The immediate treatment was given to the patient with antiplatelet agents (aspirin 300 mg), antihypertensives and insulin therapy since admission. Dizziness improved within some days in this patients.

Fifteen days later, the patient was not able to fully close the right eye, as well as having difficulty in liquids consumption. On examination,the patient had disclosed bilateral peripheral facial paralysis, including flattening of the nasolabial fold and absent forehead wrinkling bilaterally (Fig. 1G). Brain computed tomography was normal. Repeat MRI was performed at day 18 from the onset of symptoms and it showed worsening of the pontine infarct (Fig. 2A–D). Patient was treated with aspirin 100 mg and plavi × 75 mg which synergistically inhibit platelet aggregation. Acupuncture and intensive rehabilitation treatment were also received. The patient had uncontrolled arterial hypertension, hyperglycemia and severe obstructive sleep apnea hypopnea syndrome, it is possible that the exacerbation of cerebral infarction was due to ischemia and cerebral edema of pontine.

At 4 months after admission, slurred speech and dizziness disappeared, the patient's diplopia was gradually improved (Fig. 1H). The patient rejected the doctor's suggestion to perform a follow-up resonance imaging of the brain.

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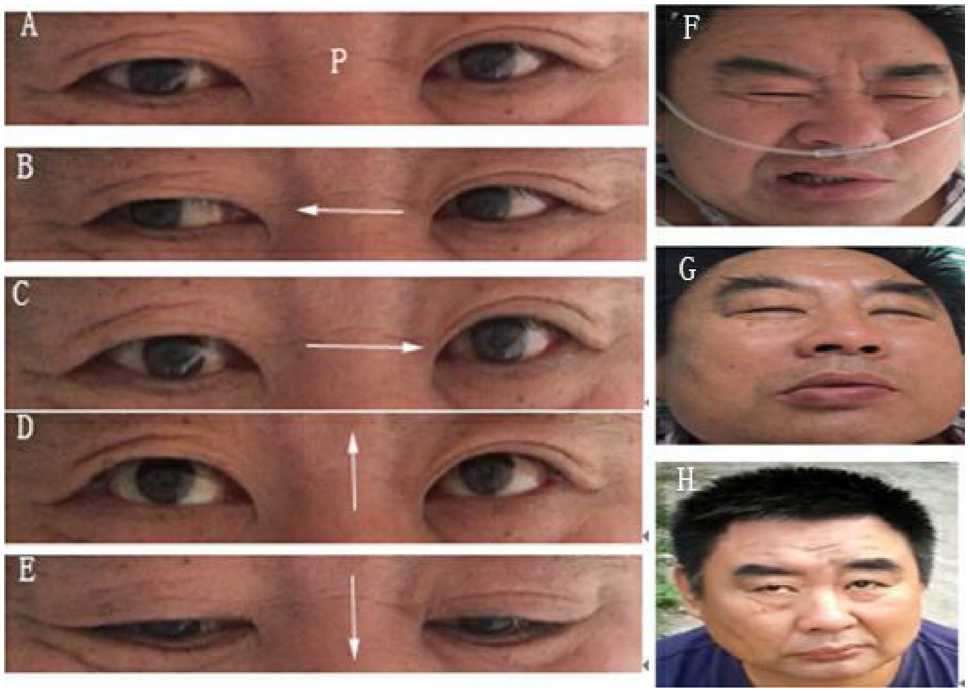

Fig. 1. The patient had a conjugate gaze palsy to the left and impaired adduction in the left eye (A–C). Noting the normal vertical eye movements from the primary position of gaze (D,E). (P- Primary position of gaze, arrows point towards the direction of gaze shifts). (F) Left lower motor neuron facial weakness after the onset of symptoms. (G) Bilateral peripheral facial paralysis occurred on the fifteenth day after admission. (H) It demonstrated resolution of peripheral facial paralysis in 4 months after admission.

3. Discussion

Although the combination of OHS and ipsilateral seventh nerve palsy, known as"eight-and-a-half syndrome", has been reported, we are unaware of an OHS with double facial nerve palsies, as our patient had. Fifteen-and-a-half syndrome, which is OHS with facial diplegia is an extremely rare clinical entity caused by a lesion of tegmentum of pons.

The structural basis of fifteen-and-a-half syndrome is damage to the ipsilateral

PPRF, bilateral MLF, abducens nucleus or fasciculus and both facial nerve fasciculi, thus the symptoms comprise of lateral gaze palsy, internuclear ophthalmoplegia and bilateral facial paralysis (Fig. 3). The etiology is not yet understood due to its extreme rarity. While the most frequent underlying causes of eight-and-a-half syndrome are infarction [3], hemorrhages [4], and demyelination [5], the fifteen-and-a-half syndrome is caused by infarction which is rarely reported [1]. Pathogenesis attributed to pontine tegmental infarct is because of occlusion of paramedian penetrating arterial branches which supply the dorsal pontine tegmentum bilaterally. For this patient we gave the antiplatelet therapy and achieved favourable outcomes, including improvement in ophthalmoplegia and peripheral facial paralysis.

4. Conclusion

Fifteen-and-a-half syndrome is a rare case of one-and-a-half syndrome plus bilateral seventh nerve palsy. Thus, the recognition of this syndrome allows precise localization of the lesion to lower pontine tegmentum. Its recognition is therefore of considerable diagnostic value.