Clinical Case Report

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Acute parkinsonism as an unexpected consequence of pituitary adenoma resection

A case report

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Abstract

Introduction: Transsphenoidal resection of pituitary tumors is a surgery performed through the nose and sphenoid sinus to remove pituitary tumors. Disorders of sodium balance are common after transsphenoidal surgery involving the pituitary gland. Here, we report the clinical features of an original case of acute onset parkinsonism later confirmed to be secondary to transsphenoidal resection of pituitary adenoma.

Patient concerns: A 36-year-old female had received transsphenoidal pituitary resection for pituitary adenoma. Eight days after the surgery, she suffered from acute onset general weakness and nausea/vomiting. She was diagnosed with hyponatremia for which she was treated. Acute onset ataxia, bilateral hand tremor, and dysarthria were then noted on the 4th day of hyponatremia treatment.

Diagnosis: Based on history, clinical manifestation, and MRI brain images, a diagnosis of acute parkinsonism caused by isolated extrapontine myelinolysis (EPM) was made.

Interventions: Patient was treated with levodopa/carbidopa.

Outcomes: Patient's symptoms and signs improved gradually and 2 month follow-up MRI brain showed significant resolution of the bilateral lentiform nuclei hyperintensities on the T2-weighted images. Her neurological deficits had subsided completely.

Lessons: This case highlights an unexpected association between transsphenoidal resection of pituitary tumors and acute parkinsonism which is a treatable manifestation of EPM. Correction of hyponatremia following transsphenoidal pituitary resections should be preceded cautiously because even gradual correction of hyponatremia can produce myelinolysis.

Abbreviations: CPM = central pontine myelinolysis, CT = computed tomography, EPM = extrapontine myelinolysis, MRI = magnetic resonance imaging, ODS = osmotic demyelination syndrome.

Keywords: extrapontine myelinolysis, parkinsonism, pituitary adenoma

1. Introduction

Osmotic demyelination syndrome (ODS) is the more recent term replacing central pontine myelinolysis (CPM) and extrapontine myelinolysis (EPM).[1] ODS could be a complication of hyponatremia correction even when the correction rate is slow.[2] Isolated EPM is rare and can present with acute onset parkinsonism. However, isolated EPM presented with acute onset parkinsonism tends to have a better prognosis when treated with levodopa/carbidopa.[3] In rare instances, hypopituitarism and hypoadrenalism can predispose to ODS in a patient with hyponatremia.[4] Herein, we describe an original case of acute onset parkinsonism later confirmed to be secondary to transsphenoidal resection of pituitary adenoma.

2. Case report

A 36-year-old Asian female presented to our emergency department due to acute onset ataxia, bilateral hand tremor, and dysarthria. At our emergency department, she was afebrile with normal pulse and blood pressure. Cardiovascular, respiratory, and abdominal examinations were all normal. Neurological examination revealed bilateral ataxia with hand tremor, dysarthria, bilateral cogwheel rigidity with left side predominance, hypokinesia, mask face, and bilateral decreased arm swings. Her cranial nerves were intact, muscle power was full but the tendon reflexes were increased bilaterally without clonus. Sensory function was also intact. The blood tests were normal including electrolytes, hepatic, and renal functions. Acute parkinsonism was diagnosed and the patient was admitted for further management. Tracing back her history, she received

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P-LH and Y-CC contributed to the work equally.

Informed written consent was obtained from the patient for publication of this case report and accompanying images.

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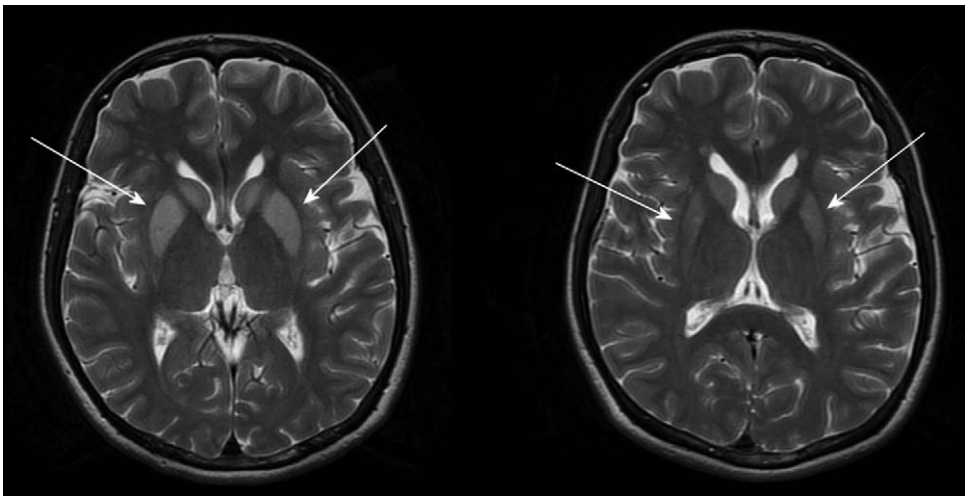

Figure 1. Left panel: Brain MRI revealed symmetric T2 hyperintensities of bilateral basal ganglia (white arrow). Right panel: Resolution of bilateral T2 hyperintensities (white arrow) 2 months later.

transsphenoidal pituitary resection for pituitary adenoma at other hospital 11 days ago and got discharged under stable condition. Eight days after the surgery, she suffered from acute onset general weakness and nausea/vomiting. She visited the hospital where she received surgery and hyponatremia 115 mmol/L was noted, she was then admitted for further management. At other hospital, 3% NaCl was given with a speed of 12 mL/h and serum sodium was corrected to 118 mmol/L on the 2nd day and 127 mmol/L on the 3rd day of admission, respectively. Acute onset ataxia, bilateral hand tremor, and dysarthria were then noted on the 4th day of hyponatremia treatment. Brain computed tomography (CT) was performed but revealed no specific finding. She then decided to get discharged and come to our hospital for further evaluation. After admission, surveys for pituitary function and thyroid function revealed normal results and the cortisol level was also normal. Brain magnetic resonance imaging (MRI) revealed symmetrical hyperintensities on the T2-weighted images over bilateral lentiform nuclei while no abnormal signals were noted over the pons, cerebellum, or other cerebral white matter regions (Fig. 1). Altogether, findings including history, clinical manifestation, and brain images, were all compatible with the diagnosis of isolated EPM. Patient's symptoms and signs improved gradually under levodopa/carbidopa treatment. Finally, only mild rigidity and dysarthria remained and the patient got discharged. Follow-up brain MRI 2 months later revealed significant resolution of the bilateral lentiform nuclei hyperintensities on the T2-weighted images. Her neurological deficits had subsided completely and she returned to normal daily life.

3. Discussion

The term "central pontine myelinolysis" was first described in alcoholic patients by Adams et al.[5] ODS can be subdivided into CPM and EPM by MRI findings involving pontine or extrapontine structures.[1] Although the exact incidence of ODS is not known, a pathology-based review article documented a prevalence rate of 0.25% to 0.5% in the past 50 years, from the time of the initial report in 1959.[6] Hyponatremia is a common complication after transsphenoidal surgery (18%), and most are asymptomatic (about 93.8%).[7] The incidence of myelinolysis following transsphenoid surgery is rare, and only a very few cases were published.[23] Patient with cardiac, renal, and thyroid disease, female patients, and patients who received postoperative cerebrospinal fluid drainage may have higher risk developing postoperative hyponatremia. There are some hypotheses that have been published to explain the possible pathophysiology of ODS. Traditionally, it was thought to be highly linked with over and rapid correction of electrolyte imbalance, especially sodium.[8–10] Now the theory of the breakdown of the blood–brain barrier has also been widely accepted. While managing patient with hyponatremia, hypertonic saline solution may lead to brain cell dehydration, and losing of electrolytes and organic osmolytes.[2] All this will induce edema and destruction of blood–brain barrier, resulting in the opening of tight junctions. The oligodendrocytes, which create the myelin sheath, seem to be vulnerable to this form of osmotic change.[11] Patients suffered from metabolic syndrome (including diabetes and hypertension), malnutrition, hypophosphatemia, renal and liver function impairment (including viral hepatitis and Wilson disease) may have higher risk of developing ODS.[12–14]

Transsphenoidal resection of pituitary tumors is a common surgical procedure to remove pituitary tumors. After operation, hyponatremia commonly occurs as a result of an abnormal sympathetic hypothalamic outflow and release of hormones, which may due to destruction of nerve conduction in the posterior pituitary gland.[15,16]

Hyponatremia is also a common cause of readmission to the hospital after the surgical procedure. Correction of hyponatremia should be preceded cautiously because even gradual correction of hyponatremia can produce myelinolysis, resulting in ODS.[17] CPM generally presents with tetraparesis and brain stem dysfunction that may include pontine dysfunction, pseudobulbar palsy and a locked-in syndrome. EPM is rare and has been described in 10% of patients with CPM. Isolated EPM is even rarer and may present as acute parkinsonism and extrapyramidal symptoms.[18,19] Thus, it may be challenging to diagnose isolated EPM in the first place.

Our patient did receive hyponatremia treatment with 3% NaCl with a speed of 12 mL/h at other hospital 8 days after operation. Although she did not have metabolic syndrome and her pituitary,

thyroid function, cortisol level were all normal, ODS still took place.

When a patient presented to the hospital with acute parkinsonism, etiologies including structural lesion (stroke, subdural hematoma, CPM, EPM, tumor, hydrocephalus), drugs (phenothiazine, thioxanthene, butyrophenone, metoclopramide, antidepressant), psychiatric (conversion disorder, obsessive compulsive disorder, malingering), infectious, postinfectious, autoimmune (systemic lupus erythematosus), and toxic (carbon monoxide, MPTP (1-Methyl-4-phenyl-1,2,3,6-tetrahydropyridine), ethylene oxide, ethanol, methanol, disulfiram) should be considered. Obtaining a brain CT image is essential for the workup of etiologies. A brain CT can readily identify structural lesions such as stroke, subdural hematoma, tumor, and hydrocephalus. However, structural lesions caused by central and extrapontine myelinolysis may be difficult to be identified by brain CT. Thus, a thorough history taking and brain MRI may be crucial to confirm the diagnosis of acute parkinsonism caused by EPM. Typically, the ODS lesions are hypointense on T1-weighted, hyperintense on T2-weighted, hyperintense on fluid-attenuated inversion recovery, and diffusion weighted imaging sequences.[1,20]

Acute parkinsonism as a consequence of EPM may respond well to dopamine supply and immunotherapy.[11,21,22] Besides, previous studies have revealed that the volume of T2 signal change on MRI is not directly correlate with clinical outcome.[22]

The above case report highlights an unexpected association between transsphenoidal resection of pituitary tumors and acute parkinsonism which is a treatable manifestation of EPM.[23] Clinicians should be aware of this unique condition as a reversible cause of acute parkinsonism.

Author contributions

Conceptualization: Chun-hsin Teng, Poyin Huang.
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Writing – review & editing: Pei-Lin Ho, Yin-Chun Chen.

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