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Editorial Board Member of *World Journal of Clinical Cases*, Ashu Seith Bhalla, MD, Professor, Department of Radiodiagnosis, All India Institute of Medical Sciences, New Delhi 110029, India

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Rehabilitation of anterior pituitary dysfunction combined with extrapontine myelinolysis: A case report

Ming-Xuan Yang, Xue-Nong Chen

ORCID number: Ming-Xuan Yang (0000-0002-5580-7624); Xue-Nong Chen (0000-0001-8414-7496).

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Ming-Xuan Yang, Xue-Nong Chen, Department of Rehabilitation, Affiliated Hospital of Zunyi Medical University, Zunyi 563003, Guizhou Province, China

Corresponding author: Xue-Nong Chen, BSc, Chief Doctor, Department of Rehabilitation, Affiliated Hospital of Zunyi Medical University, No. 113 Dalian Road, Zunyi 563003, Guizhou Province, China. ymx0019@sina.cn
Telephone: +86-15329926285

Abstract

BACKGROUND

Extramedullary myelinolysis is a rare demyelinating disease, often caused by rapid increases in serum sodium concentration in patients with hyponatremia. Clinical manifestations are neuropsychiatric symptoms, limb weakness, and dysarthria. Because of its poor prognosis and high disability rate, it poses a huge burden on the global economy, societies, and families. This article reports rehabilitation in a patient with pituitary dysfunction combined with extramedullary myelinolysis.

CASE SUMMARY

A 27-year-old Chinese man developed anorexia, vomiting, and limb weakness and was diagnosed with pituitary insufficiency. He had low serum sodium, slow movement, muscle weakness, and muscle tone abnormalities after sodium supplementation, involuntary limb shaking, ataxia, and dysarthria. According to the symptoms and signs and imaging reports, he was diagnosed with extramedullary myelinolysis. After treatment with hormone therapy and neurotrophic drugs, motor and speech function did not improve, so he was treated in the rehabilitation department for 4 wk. The patient's physical status was improved substantially during his stay at the rehabilitation department.

CONCLUSION

Patients with extramedullary myelinolysis who actively participate in rehabilitation intervention can significantly improve their activities of daily living.

Key words: Anterior pituitary dysfunction; Extrapontine myelinolysis; Rehabilitation; Motor function; Speech function; Case report

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Core tip: Medical treatment of extramedullary myelinolysis is beneficial for alleviating development of the disease, but treatment of patients with motor dysfunction, dysarthria, and other complications is poor, leading to difficulties in patient daily living, self-care, and social activity. Active intervention in rehabilitation has a beneficial effect on patient functional recovery.

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INTRODUCTION

Anterior hypopituitarism is caused by hypophyseal lesions of different types, which lead to hypophysin secretion deficiency and secondary hypofunction of thyroid and adrenal glands. Adrenocortical dysfunction may cause abnormal water and sodium metabolism and hyponatremia^[1]. Under these conditions, if serum sodium is rapidly corrected, cell metabolic disorder^[2] and osmotic demyelination syndrome may occur in the central nervous system (CNS). Usually, osmotic demyelination syndrome includes central pontine and extrapontine myelolysis. Clinical features include dysarthria, quadriplegia, dyskinesia, encephalopathy, or atresia syndrome^[3]. Generally, the prognosis of demyelinating diseases is poor and there is no effective treatment^[4]. Therefore, it is suggested that patients should receive active rehabilitation including physical therapy, occupational therapy, and speech therapy^[5].

CASE PRESENTATION

Chief complaints

A 27-year-old Chinese man with pituitary dysfunction presented with unclear speech and unstable walking for > 6 mo.

History of present illness

On February 21, 2019, the patient had oral ulcers and gradually developed anorexia, nausea, vomiting, constipation and limb weakness, pallor, and consciousness disorder. He went to the hospital for treatment, and was diagnosed with anterior pituitary hypofunction, pituitary crisis, and pituitary microadenoma. Serum sodium level was 102.7 mmol/L and he was treated with a high concentration of saline sodium. Over the next 3 d, serum sodium increased to 121.8, 134.5, and 137.7 mmol/L. Levels of sex hormones, growth hormone, and thyroid function were lower than normal. During hospitalization, the patient was treated with sodium, potassium, glucocorticoid, thyroid hormone and androgen supplementation, anti-infective agents, and gastric mucosal protective agents.

After discharge on March 2, 2019 the patient developed speech difficulties, nausea, retching, constipation, fatigue, involuntary limb shaking, poor eating, and reduced stool frequency. He was readmitted on March 12, 2019. Pituitary computed tomography (CT) showed suspicious nodular foci, and cerebral CT showed multiple low-density lesions in the bilateral basal ganglia. Cranial magnetic resonance imaging (MRI) and diffusion-weighted imaging showed abnormal signals in the bilateral caudate nucleus and globus symmetry abnormality, which suggested metabolic or toxic encephalopathy; vacuolar saddle signs; and left and upper sinus submucosal cysts. The diagnosis was anterior pituitary hypofunction, extramedullary myelinolysis, and pituitary microadenomas. During hospitalization, he was treated with glucocorticoids, thyroid hormone supplementation, and anti-infective agents, which improved neurological function and nutritional status. At the time of discharge on March 25, 2019, there was no obvious relief in the symptoms, his speech was defective, and he had involuntarily limb shaking and increased muscle tension in the limbs. After repeated hospitalization in the endocrinology and neurology departments, the above symptoms were not significantly relieved. On July 25, 2019, he was treated in the department of rehabilitation medicine.

History of past illness

Seventeen years ago, the patient showed no obvious incentive for growth and development, his growth rate slowed down compared with that of his peers, and his height was significantly shorter.

Personal and family history

The patient had no significant past history or family history.

Physical examination

The patient was conscious, spirited, of short stature, and had a female face. His speech was defective, and he did not cough when drinking water. The pupils on both sides were about 3.0 mm in diameter, and directly and indirectly sensitive to light reflection; nystagmus was absent, and tongue tremor was present. He had normal facial muscle activity, soft neck, and weak neck shrug. Manual muscle testing (MMT) scores were as follows: bilateral supraspinatus muscle: 3 points (active movement against gravity); left biceps brachii and triceps brachii muscle: 4 points (active movement against gravity and resistance); right biceps brachii and triceps brachii muscle: 3 points; muscles associated with hand grip: 3 points; muscles of the left and right waists: 4 points; left and right hip flexors: 4 points; and left and right hip extensors: 3 points. Muscle tension in the limbs was normal. There were no obvious abnormalities in the bilateral deep and shallow sensation and complex sensation. He could maintain a dynamic balance while standing, and exercise was slow. Physiological reflexes were weakened and pathological reflexes were not elicited. Activities of daily living (ADL) score was 60 points (Table 1).

Laboratory examinations

Laboratory test results were within normal limits.

Imaging examinations

MRI craniocerebral plain scan on July 29, 2019 showed bilateral white globular abnormal signal, suspected hepatic encephalopathy, hepatocellular nuclear disease, and empty sella syndrome (Figure 1).

FINAL DIAGNOSIS

Extrapontine myelinolysis and pituitary dysfunction.

TREATMENT

Rehabilitation: To promote CNS recovery by hyperbaric oxygen therapy daily (qd), to ensure adequate oxygen supply to the CNS to promote recovery of damaged nerves; provide comprehensive training for limbs qd, to increase limb muscle strength, and prevent disuse atrophy; joint treatment for limbs qd, to improve exercise and coordination function; for both hands with fine motor dysfunction, two-hand function training qd, to improve hand joint mobility, flexibility, grip, side pinch and other functions; hand exercise qd. Acupuncture treatment qd, to increase muscle strength, improve swallowing dysphonia, and promote CNS feedback to improve motor control. Popsicles were used to stimulate muscles in the mouth, low-frequency pulse electrotherapy was used to stimulate the throat muscles, and alphabet and pronunciation training was given to improve dysarthria.

Medication: Ganglioside 40 mg, *i.v.* qd; levothyroxine tablets 50 µg po qd; vitamin B1 tablets 10 mg po qd; and prednisolone acetate tablets 5 mg po qd were administered.

OUTCOME AND FOLLOW-UP

His speech was improved although still unclear, and he had no dizziness or fatigue. Mental condition was normal. Physical examination showed stable vital signs, good cooperation with examiners, and dysarthria. Ocular tremor was absent, tongue tremor was present, facial muscle activity was normal, neck was soft, and neck shrug was strong. MMT scores after treatment were all increased to 5 points except for muscles associated with hand grip with an increased score to 4 points, in which 5 points is defined as having normal muscle strength with active movement against full resistance without evident fatigue. There was no obvious abnormality in the bilateral deep and shallow sensation and complex sensation. The standing balance was

Table 1 Modified Barthel index at admission and discharge

Items	Admission	Discharge
Eating	5	10
Bathing	0	5
Grooming	0	5
Dressing	5	10
Bowel management	10	10
Bladder management	10	10
Toileting	10	10
Bed, chair, wheelchair transfer	10	15
Walking	10	15
Stairs	0	10
Total	60	100

normal. Bilateral knee reflex, Achilles tendon reflex, periosteal reflex and biceps reflex had no obvious abnormalities. Babinski sign, Brudzinski sign, Hoffmann sign, and finger test were negative. ADL score at discharge was 100 points (Table 1) (Figure 2).

DISCUSSION

Adrenocorticotrophic hormone, produced by hypophysis, stimulates the production and secretion of adrenocortical hormone. Secondary adrenal insufficiency occurs when the anterior pituitary function is impaired. When adrenal insufficiency occurs, aldosterone secretion decreases and water and sodium excretion increase. In addition, the decrease of cortisol hormone can promote the secretion of antidiuretic hormone, causing the decrease of water excretion and hyponatremia. After rapid correction of sodium, demyelination of the CNS may occur. Patients with extramedullary myelinolysis have various behavioral abnormalities such as dance-like movements, lethargy, and apathy. The extrapyramidal symptoms can be expressed as skeletal muscle loss, parkinsonism, and hand and foot movements^[2,6]. The mechanism is not completely clear at present, and the current view is that the steady-state condition of nerve cells depends on the migration of inorganic ions inside and outside the cell. When serum sodium is quickly corrected, the external conditions change. If the ion exchange of the cells does not reach equilibrium, the cells either synthesize the organic permeate or transport the ions from the external environment into the cell. Both methods need energy and time; if the extracellular tension rises too fast, the cells are not able to maintain osmotic pressure balance, resulting in damage. Oligodendrocytes are particularly susceptible to apoptosis due to the above process. Once patients have osmotic demyelination, it is usually only partially reversible^[7] and drug therapy is essential. Thyroid-stimulating hormone and corticosteroids are effective, and plasma exchange is also used to treat extrapontine myelinolysis. In addition to active drug treatment, it is equally important to improve the quality of life of patients and prevent complications such as aspiration pneumonia, deep vein thrombosis and pulmonary embolism^[8]. Therefore, such patients should actively carry out rehabilitation interventions to reduce morbidity and mortality.

Hyperbaric oxygen therapy is considered to improve blood circulation and perfusion, promote fibroblast growth, supply oxygen to damaged tissues, increase neuronal oxygen reserve, promote reversible nerve fiber regeneration and remyelination, and increase aerobic metabolism^[9]. It has a good therapeutic effect on demyelination.

In the present case, before treatment, the patient's limb movement was slow, his limbs performed daily life tasks poorly, and muscle strength was weak. He could only maintain a dynamic balance while standing. Therefore, he was treated mainly by comprehensive training for limbs, including the methods of proprioceptive neuromuscular facilitation and motor relearning program, to improve limb mobility and enhance muscle strength. After 2 wk, the patient's muscle strength was significantly improved. At that time, motor function focused on coordination ability, and joint treatment of the limbs promoted coordinated movement of hands and feet to improve balance and walking function^[10]. In addition, joint treatment of limbs improved motor function while preventing complications such as muscle atrophy, muscle contracture, and lung infection^[11,12].

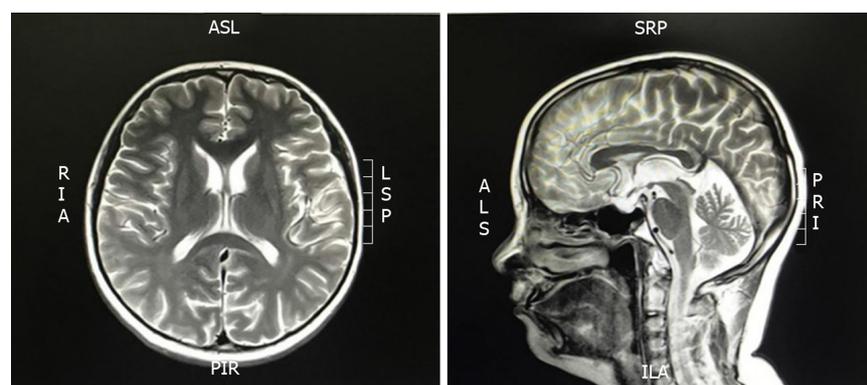


Figure 1 T2-weighted sequences showing bilateral globus pallidus abnormal signals. Hyperintense lesions in the midbrain, pons, and thalamus consistent with osmotic demyelination syndrome.

Two-hand function training and occupational therapy can improve hand function. Poor fine movement of the hands means that movement is slow. By functional training, flexibility of the hands is improved, hand-eye coordination training is emphasized, and the hand grip is improved. Occupational therapy is used to increase hand movement of patients' daily tasks, which is important for improving activities of daily living and quality of life^[13].

Acupuncture is believed to improve nerve conduction function, activate brain nerve function, increase muscle strength, and promote motor function recovery. Acupuncture protects the myelin sheath by promoting oligodendrocyte proliferation and inhibiting glial cell death^[14]. Acupuncture in the oropharynx can improve dysarthria and swallowing function.

Consonant function training, oral ice stimulation, and pulse electrotherapy are also used in patient rehabilitation. All types of dysarthria will have false vocalization. The reason is that dysfunction of the soft palate leads to insufficient oral pressure. Oral ice stimulation involves cold stimulation of oral muscle contraction, and increases muscle strength of muscles such as the tongue. Pulse electrotherapy uses electrical impulses to stimulate the throat muscles and enhance feedback. Articulation training establishes a normal pronunciation pattern using syllable training, corrects wrong articulation, and improves dysarthria.

CONCLUSION

Active rehabilitation for patients with extramedullary myelinolysis can improve the coordination and fine movement of patients, and significantly restore activities of daily living, but the recovery of speech function is slow.



Figure 2 After receiving 4 wk of treatment, the patient could put on clothes and socks independently.

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