

Reversible lesion splenial syndrome (RESLES) associated with central pontine myelinolysis: A case report

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Abstract

Reversible Splenial Lesion Syndrome is a distinct clinico-radiological syndrome of varied etiology characterized by transient lesions involving the splenium of the corpus callosum. The symptomatology isn't specific and the evolution is benign except in those patients with an underlying severe disorder. We here described a patient case with RESLES syndrome and central pontine myelolysis accompanied by acute pancreatitis and dysnatremia. Our purpose is to define this syndrome, its etiology, presentation, prognosis, and possible patho-physiological mechanisms.

Keywords

reversible splenial lesion syndrome; central pontine myelinolysis; acute pancreatitis; dysnatremia

Introduction

Reversible Splenial Lesion Syndrome (RESLES) is a disorder characterized by the presence of a focal lesion often involving the central area of the Splenium of the Corpus Callosum (SCC). Clinical presentation is non-specific and depends on etiology. It is more commonly seen among patient with encephalitis, infection, malnutrition, systemic lupus erythematosus, high-altitude cerebral edema, seizures and antiepileptic drug withdrawal, eclampsia and metabolic disturbance [1]. Neuroimaging shows a non-enhancing round-shaped lesion centered in the SCC that disappears after a variable lapse [1]. We here describe a case of patient with RESLES associated with acute Pancreatitis, dysnatremia and central pontine myelolysis. To our knowledge this is the first case report of this kind. We performed systematic research of literature to define this syndrome, its etiology and possible patho- physiological mechanisms.

Case Report

A 31 years old woman was admitted to hospital for abdominal pain, vomiting, sputum yellow color,

fever and general state alteration at 15th week's amenorrhea. Her medical history was unremarkable and was not taking any medication. On examination, the patient has dehydration, tachycardia at 120 beats/min, low blood pressure at 95/49 mmHg, Glasgow confusion 14/15, chest's examination finding snoring rattle. Laboratory findings showed acute renal failure, urea 4,9g/l and creatinine 122 mg/l GFR 3,83 ml/min/1,73m², an elevated lipase 1150 UI/l, elevated C-reactive protein 68 mg/l and hyponatremia 118 mmol/l. Abdominal and pelvic ultrasound has been done in first line showing alithiasis distended gall bladder, non-evolutive pregnancy without cardiac activity, normal kidney's morphology. X-ray of chest revealed a pulmonary infection. Initial management consisted in rehydration with antibiotic cephalosporin 3rd generation and fetal expulsion was indicated. A few days after, the patient developed diplopia, decreased visual acuity and gait disturbance with persistence vomiting. A second laboratory finding showed hypokalemia 2,2 mmol/l, hypernatremia 154 mmol/l, elevated lipase 779 UI/L with normal renal function. An abdominal CT complement showed pancreatitis C stage according to Balthazar's classification. fundoscopy showed bilateral papillary edema stage II confirmed by OCT. Initial MRI of the brain revealed a lesion involving the splenium of the corpus callosum measured 16*16*16 mm isosignal on T1, intermediate signal on T2 and flair hyper-signal diffusion with restriction ADC (apparent diffusion coefficient) and not raised after contrast injection. Presence of hypersignal T2 centro-pontic not enhanced after contrast injection. It is not surrounded by peri-lesional edema and has no mass effect on near structures. No abnormality in the MRI angio sequence (Figure 1).

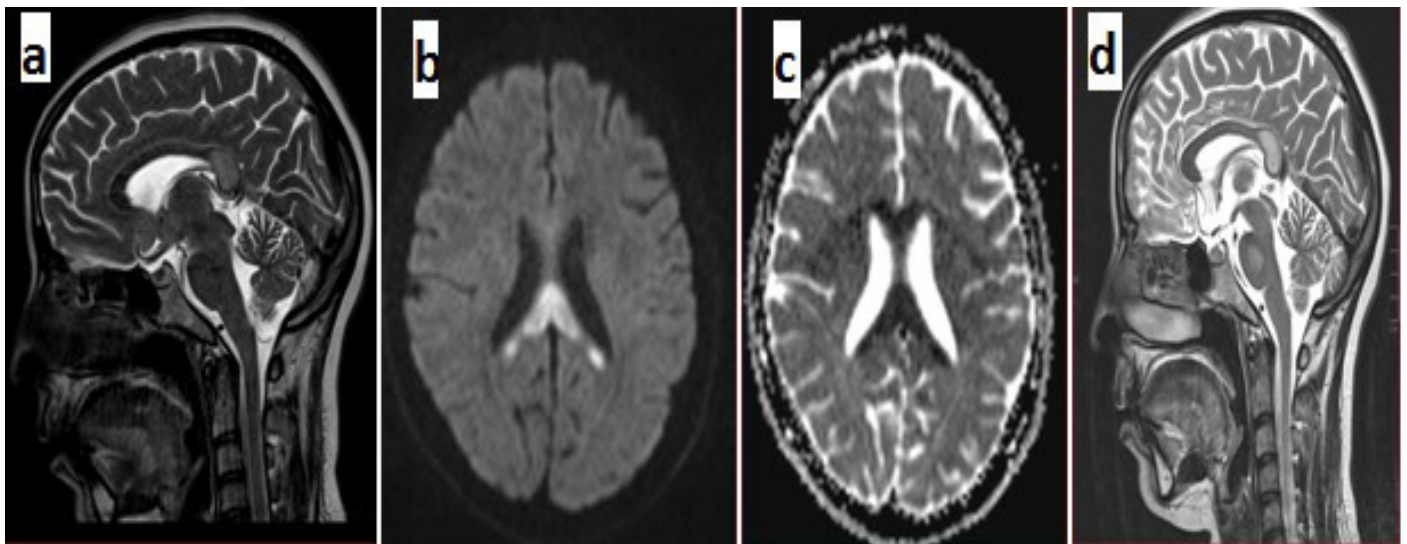


Figure 1: MRI Imaging of the brain on admission with (a) T2 sagittal, (b) axial diffusion, (c) ADC, (d) T2 sagittal on third day. A lesion involving the splenium of the corpus callosum measured 16*16*16 mm intermediate signal on T2 and flair hyper-signal diffusion with restriction ADC. Presence of hypersignal T2 centro-pontic not enhanced after contrast injection

The patient was rehydrated with serum glucose 5%, she was kept fasting and a correction of hypokalemia and hypernatremia was introduced.

After 3 weeks the patient presented with complete clinical recovery and normalization of visual acuity. A follow-up MRI one month after first MRI demonstrated normalization of the signal within the splenium of the corpus callosum without any cerebral anomaly (Figure 2).

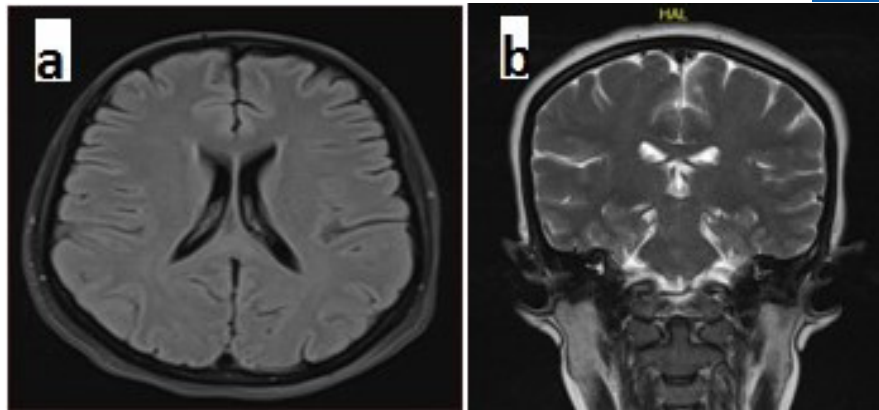


Figure 2: MRI Imaging of the brain after one month demonstrated normalization of the signal within the splenium of the corpus callosum without any cerebral anomaly. (a) flair axial, (b) T2 coronal.

Discussion

RESLES is a disorder involving transient lesions in the SCC. It was first reported by Kim and colleagues [2] in 1999, describing a group of epileptic patients with concurrent corpus callosum lesions, deducing that these lesions may be caused by the use of the antiepileptic drug. Since then, there have been other cases of reversible lesions of the corpus callosum reported including infections, metabolic conditions, other pharmacological agents, anorexia nervosa, malnutrition, vitamin B12 deficiency, Charcot-Marie-Tooth disease, high-altitude cerebral edema, systemic lupus erythematosus, and eclampsia [3].

The pathophysiology of these reversible splenium lesions of the corpus callosum is not very clear. Many RESLES cases shows data supporting that intramyelinic cytotoxic edema plays an important role, as inferred by diffusion-restriction and low Apparent Diffusion Coefficient (ADC) values on Diffusion-Weighted Imaging (DWI) [4]. Level fluctuations combined with changes in salt homeostasis and resultant myelin edema are other suggested mechanisms [4].

In our case reported, the patient has acute pancreatitis with vomiting and dehydration accompanied by a mild hyponatremia. And she has two rare disease central pontinemyelinolysis associated with RESLES syndrome. Therefore, to our knowledge, we are the first to report this association case.

Central Pontinemyelinolysis (CPM) is neurological disorders characterized by demyelination in the pons. Hyponatremia and rapid correction of sodium blood-concentration are known risk factors for developing CPM [5]. This raises the question, whether hyponatremia is a part of pathophysiology of RESLES as CPM or only an accompanying circumstance.

Interestingly, similar MRI findings as are typically described in RESLES can be shown in several patients with osmotic demyelination syndromes like Central Pontinemyelinolysis (CPM) or Extrapontinemyelinolysis (EPM) [5]. In CPM/EPM rapid improvement of ADC values predicts a good clinical recovery [6], which is also characteristic in RESLES. In this sense, the reversible lesion in the SCC as seen in our patient can be explained by dysnatremia.

Conclusion

RESLES is a reversible condition in most patients, clinically and radiologically. The association with CPM suggested Weather RESLES and CPM have a similar pathophysiology or RESLES is a specific type of CPM remains to be elucidated in future studies.

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