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A case of reversible splenial lesion syndrome in early post-partum period☆

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1. Introduction

Since was first described in 1999 by Kim et al. [1] reversible Splenial lesion syndrome (RELEs) is an increasingly Recognized Syndrome of unknown etiology defined as reversible isolated splenial lesions in the corpus callosum, Readily Identified by Magnetic Resonance Imaging (MRI) and Typically resolve completely within a short period.

RELES was described in patients with various diseases including infectious, metabolic and in patients in seizure disorders [2]. However, up to our knowledge, RELES was described in one case of post-partum preeclampsia [4].

RELES is defined as transient, well-circumscribed lesion located in the median aspect of the splenium of the corpus callosum (SCC). It is usually non-contrast enhancing on T2 and flair, associated with diffusion restriction and low apparent diffusion coefficient (ADC) values indicating Cytotoxic edema, in rare Cases, ADC is high indicating Vasogenic edema [3].

We are presenting a case of post-partum associated RELES, occurred after normal vaginal delivery, with MRI findings consistent with splenial Cytotoxic edema. Keeping this rare correlation in mind helps clinicians avoid unnecessary expensive diagnostic procedures and therapeutic interventions, which makes presenting this association highly valuable.

2. Case report

A 29-year-old female patient with past medical history significant for Anxiety, presented two weeks post non-complicated normal vaginal delivery with a one-week history of gradually progressing sever band like throbbing headache, that worsens upon standing and associated with episodes of transient visual. Patient mentioned having loss of taste sensation too. The patient denied having seizures, awaking from sleep with tongue biting or having urine incontinence. No problems with breastfeeding, or excessive vaginal blood loss.

On examination, the patient was Normotensive, afebrile, normal Heart rate, normal respiratory rate, and saturation.

The Respiratory and cardiovascular examinations were normal, the abdomen was soft and lax, no masses, and no lower limb edema was identified.

She was awake, alert, attentive, oriented to time, place and persons. Recent and remote memories were intact with no evidence of language dysfunction. No dysarthria, no anomia, and repetition was intact. The patient had satisfactory fund of knowledge as well as normal attention span.

Visual field was intact to finger counting; No neglect was identified. No papilledema or hemorrhage on the fundoscopic exam, Pupils are symmetrical and reactive, extraocular movements are intact, no nystagmus, the Facial sensation is intact bilaterally in V1-V3 distributions, no facial asymmetry, the hearing was intact to finger rub, the palate rises symmetrically, Sternocleidomastoid power is intact bilaterally and the tongue protrudes in the midline. The motor exam revealed normal tone with no resting, postural or intention tremors, Muscle strength was 5/5 in all muscle groups. Reflexes were 2+ symmetrical in biceps, brachioradialis, patellar, 1+ ankle jerks, with no clonus, toes downgoing bilaterally. No Hoffman signs, the Sensory exam was intact to light touch, pinprick, vibration, and temperature. Finger-nose-finger and heel to shin tests were normal bilaterally, rapid alternating movements were intact. There was no discernible ataxia. Stable when standing with feet together with eyes open and mild sway when closed. The gait was hesitant, with standby assist, but not shuffling or wide-based, no ataxia noted. No evidence of orthostatic blood pressure changes.

Electrolytes were normal, renal and liver functions were normal. Complete blood count showed Normal White blood cells (WBCs) with

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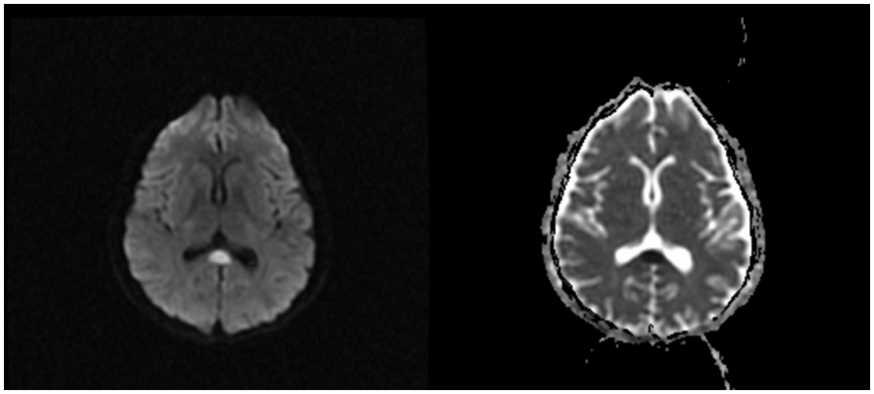

Fig. 1. Upon Presentation: Axial Diffusion-weighted imaging (DWI, B100 tensor on the left and ADC map on the right) demonstrates high signal within the splenium of the corpus callosum with loss of signal on ADC.

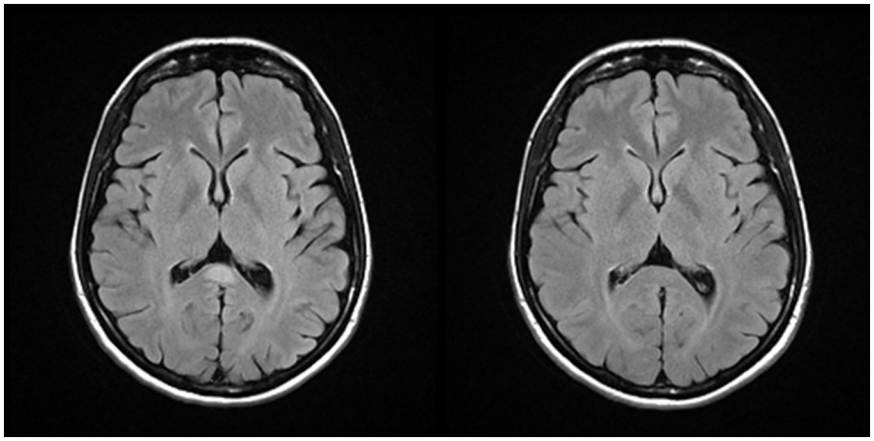

Fig. 2. T2 FLAIR images at an initial presentation on the (left) demonstrates high signal within the splenium of the corpus callosum. This corresponds to the high signal on DWI. The splenium is mildly expanded. On follow up on the (right), there is a resolution of the high signal within the splenium and return to more normal size.

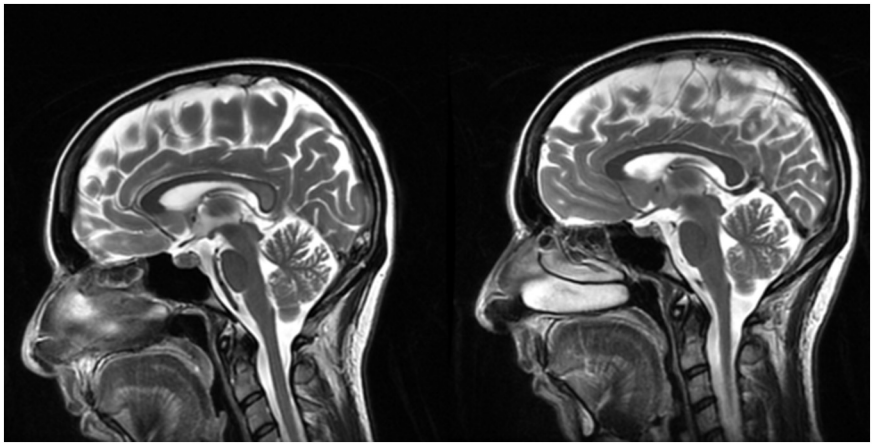

Fig. 3. Sagittal T2 weighted images at presentation (left) the midline demonstrates an expansion of the splenium of the corpus callosum with high signal seen extending into the dorsal body of the corpus callosum and on follow up (right) demonstrate resolution of high signal within the splenium of the corpus callosum and return to more normal size.

normal differential, normal hemoglobin and normal platelet count.

Cerebrospinal fluid was clear, no WBCs, normal glucose and normal protein, no organisms seen and cultures were negative, CT head rolled out intracranial hemorrhage, transcortical infarct, or mass lesion, sinuses were visualized and were normal.

Brain Magnetic Resonance Venography (MRV) rolled out cerebral venous thrombosis, however, showed restricted diffusion in corpus callosum splenium indicating Cytotoxic edema, with the possibility of Reversible splenium lesion syndrome (RELES) was raised (Figs. 1–3).

Our patients treated conservatively with Intravenous fluids, pain control and kept on IV dexamethasone. Her condition improved dramatically. Three days later the patient was discharged home.

Four weeks later the patient was seen in the clinic, her symptoms were completely resolved except for a minimal headache, the Neurologic examination was completely normal. Repeated MRI of the brain showed complete resolution of the restricted diffusion within the splenium of the corpus callosum which is compatible with RELES (Figs. 1–3).

3. Discussion

Reversible Splenial lesion syndrome (RELES) is a distinct clinical-radiological diagnosis, which means that it is diagnosed when a certain radiologic criterion is met and the clinical course is consistent with the disease expected course. RELES is of unknown pathophysiology. It is typically a benign condition and resolves spontaneously within a short

period unless the underlying disease is serious and was not treated appropriately.

We are reporting a new case of postpartum RESLES presented two weeks post normal vaginal delivery following uncomplicated pregnancy, presenting with a headache, decrease in visual acuity and loss of taste sensation. Although more reports of RESLES have been recently reported in association with postpartum preeclampsia [4], seizures and central CNS infections [2]; up to our knowledge, our case is the first reported RESLES associated with a normal peripartum period.

RESLES was also extensively reported in relation to certain conditions such as antiepileptic drug toxicity or withdrawal, high altitude cerebral edema, Machiafava-bignami disease, hypoglycemia or hypernatremia. It was also specifically reported in pediatric patients who were diagnosed with mild encephalopathy with a reversible splenial lesion in association with various possible etiologies [5].

Magnetic Resonance Venography (MRV) rolled out cerebral venous thrombosis, however, it showed restricted diffusion in the splenium of the corpus callosum raising RESLES as a possible alternative diagnosis.

The MRV Showed a well-circumscribed lesion located in the median aspect of the splenium of the corpus callosum (SCC). It was non-contrast enhancing on T2 and flair, associated with diffusion restriction and low apparent diffusion coefficient (ADC) values indicating Cytotoxic edema, which was consistent with previously reported cases [3]. The complete resolution of the splenial lesions supports the RELES diagnosis.

Our workup rolled out associated meningitis or encephalitis, also there was no evidence preeclampsia.

Keeping this rare correlation in mind helps clinicians provide reassurance to the patient and their family, although workup for metabolic and infectious etiologies in such cases remains essential.

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