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Case Report

A Rare Cause of Reversible Splenial Lesion Syndrome (RESLES): Benign Convulsions with Mild Gastroenteritis

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ABSTRACT

Transient lesions involving the splenium of corpus callosum is defined as reversible splenial lesion syndrome (RESLES). Benign convulsions with mild gastroenteritis is a rare condition which may be associated with RESLES. Since the prognosis is excellent, the awareness of this association is important to prevent unnecessary investigations and anti-epileptic drug therapy.

Keywords: Splenium, corpus callosum, gastroenteritis, seizure, RESLES

Introduction

Reversible splenial lesion syndrome (RESLES) is a rare clinico-radiological entity characterized by a transient lesion in the splenium of corpus callosum (1). RESLES may result from various causes such as infections, metabolic derangements, high-altitude cerebral edema, seizures or antiepileptic drug withdrawal. In the paediatric population, the most common form of RESLES is mild encephalitis/encephalopathy with a reversible splenial lesion (MERS) which may occur during infectious diseases (2). In MERS, encephalopathy lasts longer than 12 hours and no evidence of inflammation is found in the cerebrospinal fluid. Disturbance of consciousness lasting less than 12 hours or an absence of altered mental status during the clinical course is defined as non-MERS form of RESLES (1).

Benign convulsions with mild gastroenteritis (CwG) was first described by Mooroka in 1982. It is characterized by; (1) occurrence in previously healthy infants or young children aged between 6 months and 3 years; (2) afebrile generalized convulsions sometimes seen in clusters related to gastroenteritis without moderate to severe dehydration; (3) normal laboratory examination including electrolytes, blood glucose and cerebrospinal fluid; (4) normal interictal electroencephalography (EEG); and (5) excellent seizure and developmental outcomes (3). Rotavirus, norovirus and other round-shaped viruses are the most common causative agents related to this entity (4).

Case Report

A previously healthy 42-month-old girl with diarrhoea and vomiting for two days was admitted to our hospital

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due to a brief afebrile generalized tonic-clonic seizure. Her past medical history and family history were unremarkable. On admission, she was alert and had normal physical and neurological examination. During the emergency room follow-up, she had another afebrile generalized tonic-clonic seizure with a duration of 3 minutes and rapidly regained consciousness. Laboratory studies including hemogram, liver and kidney function tests, uric acid, serum electrolytes and acute phase reactants were normal. Stool sample analysis revealed no pathogens. EEG was found to be normal. Cranial magnetic resonance imaging (MRI) revealed increased T2-signal intensity in the splenium of corpus callosum and hypo-intense signal on apparent diffusion coefficient images (Figure 1). She had no further seizures and was discharged on the third day of hospitalization with no antiepileptic drug treatment. The follow-up cranial MRI of the patient performed after 4 weeks showed complete resolution of the lesion (Figure 2). The patient is now seizure free for 6 months with a normal neurological examination and developmental milestones.

Discussion

In 2007, Natsume et al. (5) first described the association of RESLES and CwG in two paediatric cases. After this initial description, a few cases of different ethnic origins, mainly from Asia, were published (6-8) (Table I). In a multicentre study conducted by the Tokai Pediatric Neurology Society, the frequency of RESLES in CwG was presented as being 22% (9). Kato et al. (8) reported nine Japanese cases with a transient splenial lesion of corpus callosum occurring during rotavirus gastroenteritis. While eight of these cases presented with encephalopathy, and were classified as MERS, only one case had clinical features consistent with the non-MERS form of RESLES due to CwG. In another study among 233 patients with RESLES, five patients with no clinical manifestations of encephalitis such as delirium or an altered level of consciousness were diagnosed with RESLES secondary to CwG. Acute seizure treatment with anticonvulsive drugs such as diazepam or phenobarbital, anti-viral agents and rehydration was administrated if necessary in the acute period, but no long-term antiepileptic treatment was given (7). Similar to these cases in the literature, the present case had only two brief generalized seizures without any encephalopathic features during a gastrointestinal infection. Although there is no specific laboratory finding for the diagnosis of CwG, Yoo et al. (10) found that serum uric acid levels are significantly higher in CwG patients than in patients with acute gastroenteritis and febrile seizures. However, the uric acid level of the present case was found to be within the normal range. The presence of a splenial lesion detected on the cranial MRI performed for the recurrent afebrile seizures led to the diagnosis of non-MERS form of RESLES secondary to CwG. No long-term antiepileptic therapy was given. The complete resolution of the splenial lesion as seen in the follow-up MRI, normal neurological development and the absence of recurrent afebrile seizures confirmed the diagnosis.

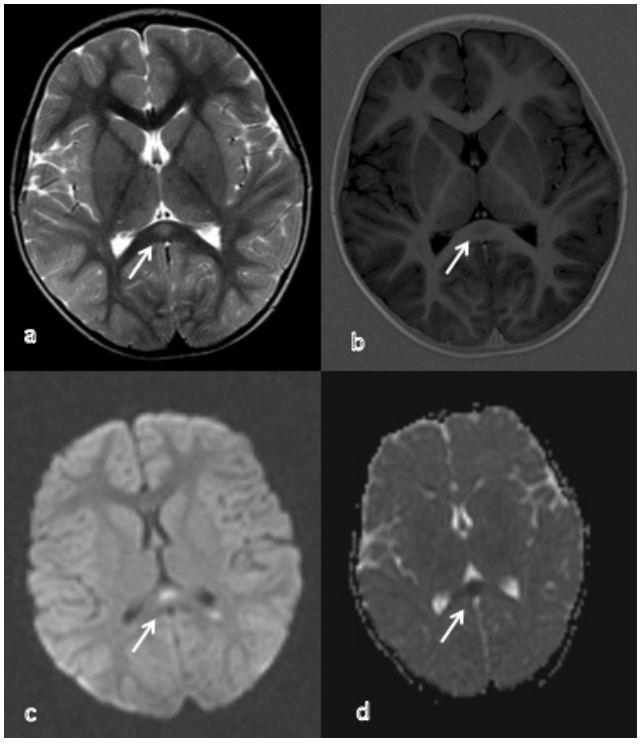

Figure 1. Cranial magnetic resonance images on admission demonstrating a splenial lesion of the corpus callosum. Axial T1-weighted (a), axial T2-weighted (b), diffusion-weighted (c), and the corresponding apparent diffusion coefficient map (d)

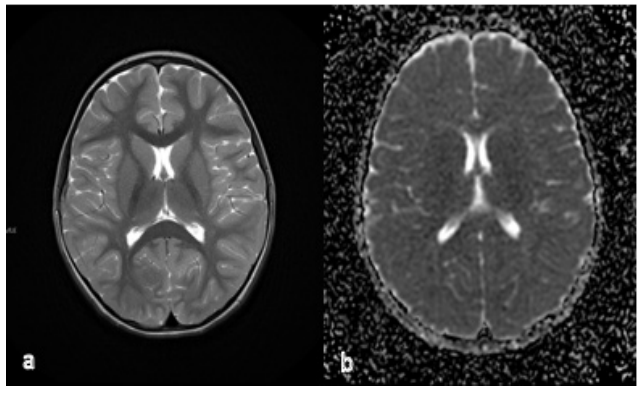

Figure 2. Follow-up cranial magnetic resonance images demonstrating the resolution of the splenial lesion. Axial T2-weighted (a) and apparent diffusion coefficient map (b)

Table I. Data of the published cases with non-MERS form of RESLES secondary to CwG

Publication	Number of patients	Age-months	Symptoms	Etiology	EEG	Treatment	Day of normal follow-up MRI/Outcome
Natsume et al. (5) 2007	2	24 36	5 episodes of focal seizures 2 episodes of generalized seizures	Rotavirus -	Normal	Diazepam, phenobarbital -	9/good 21/good
Kato et al. (8) 2009	1	12	3 episodes of tonic seizures	Rotavirus	Intermittent spikes, occipital slow wave	Diazepam	6/good
Jang and Lee (6)2010	1	30	2 episodes of generalized tonic-clonic seizures	Rotavirus	Normal	Rehydration, empiric antibiotics and acyclovir	6/good
Jiang et al. (7)2014	5	36 14 28 30 24	4 episodes of generalized tonic-clonic seizures 3 episodes of generalized tonic-clonic seizures 5 episodes of generalized tonic-clonic seizures 2 episodes of generalized tonic-clonic seizures 4 episodes of generalized tonic-clonic seizures	- - - - Rotavirus	Normal Normal Normal Normal Occipital slow wave	Rehydration, ribavirin, diazepam Rehydration, ribavirin, phenobarbital Rehydration, ribavirin, phenobarbital Rehydration, ribavirin, diazepam Rehydration, ribavirin, diazepam, phenobarbital	11/good 10/good 15/good 10/good 12/good
Presented case	1	42	2 episodes of generalized tonic-clonic seizures	-	Normal	Rehydration	30/good

MERS: Mild encephalitis/encephalopathy with a reversible splenial lesion, CwG: Convulsions with mild gastroenteritis, RESLES: Reversible splenial lesion syndrome, EEG: Electroencephalography, MRI: Magnetic resonance imaging

Conclusion

In young children with recurrent afebrile seizures and gastroenteritis without fever, dehydration and ion imbalance, the association of RESLES and CwG should be kept in mind. Since the prognosis is excellent with no permanent neurological sequelae, awareness of this rare clinico-radiological entity is important in order to avoid unnecessary investigations and long term anti-epileptic drug therapy.

Ethics

Informed Consent: The consent form was received from the parents of the patient.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Concept: N.U., A.D., S.K., S.Y., Design: N.U., A.D., S.K., S.Y., Data Collection or Processing: N.U., A.D., S.K., S.Y., Analysis or Interpretation: N.U., A.D., S.K., S.Y., Literature Search: N.U., A.D., S.K., S.Y., Writing: N.U., A.D., S.K., S.Y.

Conflict of Interest: No conflict of interest was declared by the authors.

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