ACADEMIC JOURNAL OF HEALTH

CASE REPORT

Reversible Splenial Lesion in Children: A Two-Case Report

ABSTRACT

"Reversible splenial lesion syndromes" (RESLES) is a rare and complex syndrome, involving diseases or conditions characterized by an isolated reversible lesion in the splenium of the corpus callosum with a transient and abnormal diffusion restriction detected by magnetic resonance imaging (MRI). The pathogenesis of this syndrome is unclear, and the prognosis is good. It can often present with a clinically mild encephalitis/encephalopathy with a reversible splenial lesion (MERS) in children. This study presents two cases of RESLES in 16-year-old and 5-year-old male patients who were found to have abnormal diffusion restriction in the splenium on MRI and presented with two different clinical presentations. RESLES and MERS should be kept in mind in the differential diagnosis of acute encephalitis/encephalopathy and nonspecific neurological symptoms.

Keywords: Children, encephalopathy, mild encephalitis, reversible splenial lesion syndromes

"Reversible splenial lesion syndromes" (RESLES) is a rare syndrome in children, involving diseases or conditions characterized by an isolated reversible lesion in the splenium of the corpus callosum (SCC) with a transient and abnormal diffusion restriction detected by magnetic resonance imaging (MRI) (1). RESLES is a complex condition reflecting various diseases, and it can often present with a clinically mild encephalitis/encephalopathy with a reversible splenial lesion (MERS) in children (2). Here, we describe two cases of RESLES in 16-year-old and 5-year-old male patients who were found to have abnormal diffusion restriction in the splenium on MRI and presented with two different clinical presentations.

CASE 1

A previously healthy 16-year-old male was admitted to the pediatric emergency department (PED) with complaints of fever, severe headache, weakness, and cramps in his fingers. He described the headache as a thunderclap headache he had never experienced before. The patient was hemodynamically stable on arrival, and GCS score was 15. On physical examination, hyperventilation and carpopedal spasm were observed. There was no focal neurologic deficit. In laboratory results, we found respiratory alkalosis in blood gas and hypocalcemia in biochemistry results. The patient was given symptomatic treatment for upper respiratory infection and treatment for hypocalcemia. Cranial computed tomography (CT) was performed at the 5th hour of follow-up and reported as normal. The patient's symptoms resolved, and he was discharged. EEG monitoring and cranial and diffusion MRI were performed on the 2nd day of follow-up. No epileptic activity was observed in EEG monitoring. On MRI, a hyperintense, diffusion-restricted nodular signal intensity change was observed in the splenium of the corpus callosum in T2/FLAIR series (Figure 1A). There was no additional neurological deficit in the follow-up one month later.

CASE 2

A previously healthy 5-year-old male was admitted to the PED with fever, slowness in speech, imbalance in gait, intermittent dullness, and unresponsiveness. The patient was hemodynamically stable on arrival. GCS score was 14 (M6 V4 E4). He had flu-like symptoms, slow and meaningless speech, unbalanced gait, and his cerebellar tests were clumsy. The patient was given symptomatic treatment (hydration and analgesia) for flu-like symptoms. Cranial and diffusion MRI were performed at the 6th hour of follow-up. A hyperintense

Muhammed Mustafa Güneylioğlu[1]
Raziye Merve Yaradılmış[1]
Orkun Aydın[1]
Betül Öztürk[1]
Bilge Akkaya[1]
Ayşe Kaçar Bayram[2]
Ali Güngör[1]
Nilden Tuygun[1]

[1]*Department of Pediatric Emergency Care, Ankara Etlik City Hospital, Ankara, Türkiye*
[2]*Department of Pediatric Neurology, Ankara Etlik City Hospital, Ankara, Türkiye*

Corresponding author:
Raziye Merve Yaradılmış
✉ karaomermerve@hotmail.com

Received: January 28, 2024
Revisioned: March 08, 2024
Accepted: March 21, 2024

Cite this article as: Güneylioğlu MM, Yaradılmış RM, Aydın O, Öztürk B, Akkaya B, Bayram AK, et al. Reversible splenial lesion in children: A two-case report. Acad J Health 2024;2(2):61-63.

DOI: 10.14744/ajh.2024.69775

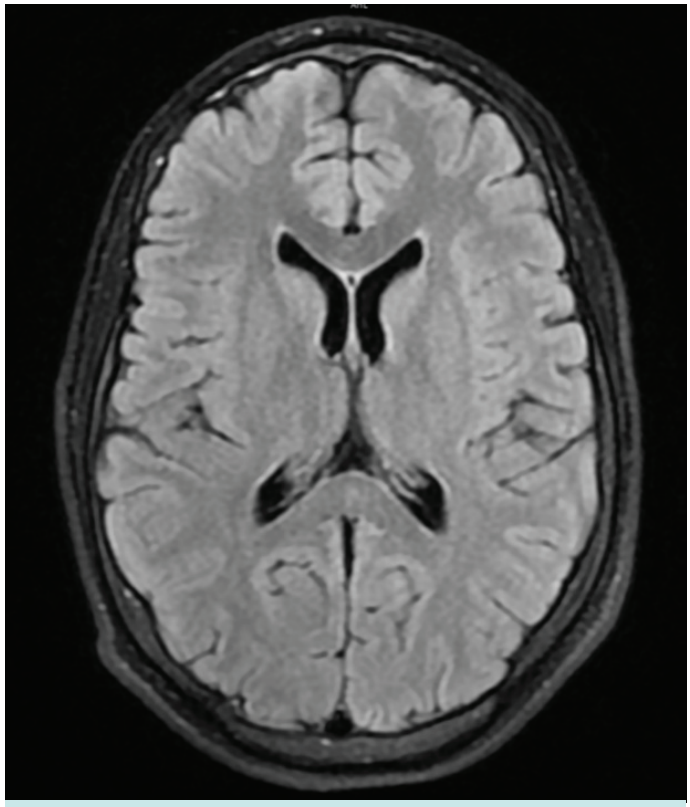

Figure 1A. Brain MRI in RESLES. A hyperintense, diffusion-restricted nodular signal intensity change was observed in the splenium of the callosum in T2/FLA series.

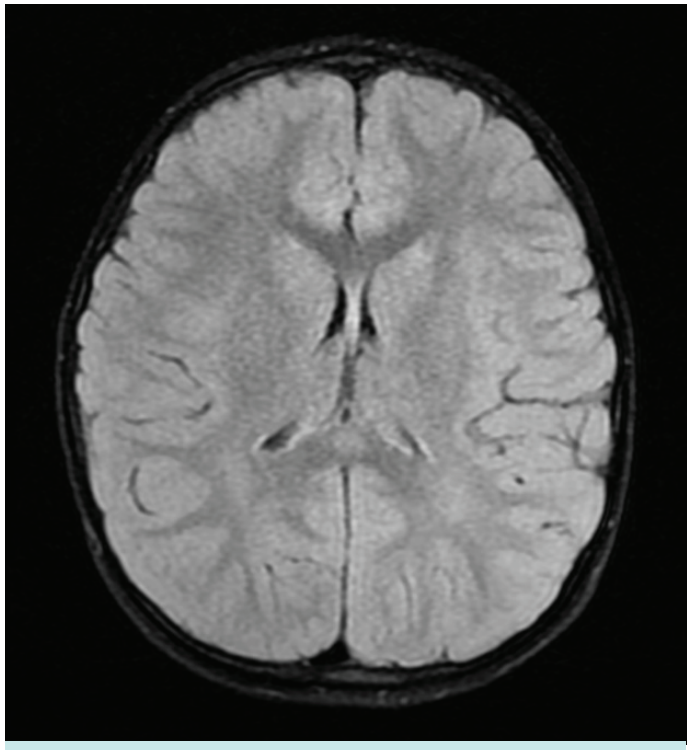

Figure 1B. Brain MRI in MERS. A hyperintense area was observed on T2/FLAIR series, showing restricted diffusion partially extending to the fornix in the middle part of the splenium.

area was observed on T2/FLAIR series, showing restricted diffusion partially extending to the fornix in the middle part of the splenium (Figure 1B). Clinical improvement was observed at the 20th hour of hospitalization. According to MRI findings, this clinical course was diagnosed as mild encephalopathy with a reversible splenial lesion associated with viral infection. There was no neurological deficit in the follow-up one month later.

DISCUSSION

This study presents two pediatric cases with different clinical features and were diagnosed with RESLES. This syndrome is a rare and complicated clinical-radiological syndrome of unclear pathogenesis, and it has a good prognosis (3). In recent years, reversible lesions especially involving the SCC have been associated with various disorders such as MERS, viral infections, high-altitude cerebral edema, metabolic disorders, seizures, antiepileptic drug withdrawal (1). The main pathogens of MERS are influenza virus (A and B), mumps virus, adenovirus, and rotavirus, although rarely bacterial pathogens (such as Streptococci, Escherichia coli, and Klebsiella pneumoniae) have also been reported (4,5). Children with multisystem inflammatory syndrome associated with severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) who developed RESLES during the course of the disease have also been reported in the literature (6).

The clinical presentation of RESLES is variable, and patients may present with nonspecific prodromal and neurological symptoms. Although the diagnosis rate has increased with the widespread use of MRI, the true prevalence of RESLES and MERS in children is still unknown (3). Fever was the most common nonspecific prodromal symptom, and its incidence in the literature was examined as 100%, 68.8%, and 57.1% (3,7,8). It may also present with signs and symptoms such as viral infection symptoms (respiratory or digestive system symptoms), parotitis, mesenteric lymph enlargement, joint effusion (3,9). Signs of altered consciousness such as drowsiness, confusion, lethargy, stupor; behavioral changes such as irritability, lags in response, impulsive behavior; motor deterioration such as muscle weakness are common neurological findings. Patients may present with seizures, headache, sensory disturbances, myoclonus, and more rarely ataxia and Babinski positivity (3,10). Clinical symptoms of case one were fever, severe headache, weakness, and cramps in fingers. He described his headache as a thunderclap headache, and nonsteroidal anti-inflammatory drug treatment did not provide relief; moreover, hyperventilation triggered by severe pain caused blood gas and electrolyte anomalies that would cause carpopedal spasm. With these findings, intracranial hemorrhage was considered as a preliminary diagnosis in the patient, and CT was performed, and it was normal. In case two, besides flu-like symptoms, there were transient altered consciousness and slowness in speech. RESLES and MERS should be kept in mind in the differential diagnosis of acute encephalitis/encephalopathy and nonspecific neurological symptoms.

Neuroimaging technique and time to imaging are determined by clinical presentation at admission and hospital conditions. Cranial CT may be preferred in the acute period to exclude intracranial pathologies, as in case one, but it does not make a diagnosis for RESLES. MRI stands out as the neuroimaging technique of choice, mainly in the SCC, or extra lesions of SCC, to detect early changes and to help interpret the nature of the lesion (cytotoxic edema, limitation in diffusion-weighted images, homogeneous and reversible features) (2,3).

CONCLUSION

RESLES and its sub-title MERS is a reversible condition with a good clinical and radiological prognosis. The true incidence of RESLES or MERS is still unknown, but as MRI facilities improve, diagnosis may increase. In this regard, it is also important to increase the awareness of RESLES for clinicians.

Informed Consent: Written informed consent was obtained from the patient's family for publication of this case report and any accompanying images.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – M.M.G.; Design – R.M.Y.; Supervision – A.G.; Resource – A.K.B.; Materials – A.K.B.; Data Collection and/or Processing – R.M.Y.; Analysis and/or Interpretation – A.G., B.A.; Literature Review – B.Ö., O.A.; Writing – M.M.G., R.M.Y; Critical Review – N.T.

Conflict of Interest: The authors declare that they have no conflict of interest.

Funding: The authors declared that this study has received no financial support.

REFERENCES

1. Garcia-Monco JC, Cortina IE, Ferreira E, Martínez A, Ruiz L, Cabrera A, et al. Reversible splenial lesion syndrome (RESLES): What's in a name? J Neuroimaging 2011;21(2):e1-14. [CrossRef]
2. Tuscano A, Zoppo M, Canavese C, Cogoni M, Scolfaro C. Transient blindness associated with mild encephalitis/encephalopathy with a reversible splenial lesion (MERS): A case report and review of literature. Ital J Pediatr 2020;46(1):1-7. [CrossRef]
3. Chen WX, Liu HS, Yang SD, Zeng SH, Gao YY, Du ZH, et al. Reversible splenial lesion syndrome in children: Retrospective study and summary of case series. Brain Dev 2016;38(10):915-27. [CrossRef]
4. Avcu G, Kilinc MA, Eraslan C, Karapinar B, Vardar F. Mild encephalitis/encephalopathy with reversible splenial lesion (MERS) associated with streptococcus pneumoniae bacteraemia. J Infect Public Health 2017;10(4):479-82. [CrossRef]
5. Uysal N, Dezhakam A, Kanmaz S, Yılmaz S. A rare cause of Reversible Splenial Lesion Syndrome (RESLES): Benign convulsions with mild gastroenteritis. J Pediatr Res 2021;8(3):359-62. [CrossRef]
6. Bektaş G, Akçay N, Boydağ K, Şevketoğlu E. Reversible splenial lesion syndrome associated with SARS-CoV-2 infection in two children. Brain Dev 2021;43(2):230-33. [CrossRef]
7. Ka A, Britton P, Troedson C, Webster R, Procopis P, Ging J, et al. Mild encephalopathy with reversible splenial lesion: An important differential of encephalitis. Eur J Pediatr Neurol 2015;19(3):377-82. [CrossRef]
8. Li C, Li C, Yang J. The clinical characteristics of children with splenium of corpus callosum solitary lesions displayed on MRI. Chin J Neurol 2014;47(12):852-56.
9. Tada H, Takanashi J, Barkovich AJ, Oba H, Maeda M, Tsukahara H, et al. Clinically mild encephalitis/encephalopathy with a reversible splenial lesion. Neurology 2004;63(10):1854-58. [CrossRef]
10. Kashiwagi M, Tanabe T, Shimakawa S, Nakamura M, Murata S, Shabana K, et al. Clinico-radiological spectrum of reversible splenial lesions in children. Brain Dev 2014;36(4):330-6. [CrossRef]