CASE REPORT OLGU SUNUMU

# A Pediatric Case of Reversible Splenial Lesion Syndrome Associated with SARS-CoV-2

## SARS-CoV-2 ile İlişkili Pediatrik Bir Reversibl Splenial Lezyon Sendromu Vakası

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**ABSTRACT** Reversible splenial lesion syndrome is characterized by a reversible lesion in the middle of the corpus callosum's splenium. This syndrome may be associated with various illnesses, including infections. For instance, a girl presented with dizziness, ataxia, and blurred vision, and was diagnosed with a reversible splenial lesion during her illness. A lumbar puncture revealed a cerebrospinal fluid pressure of 39 cm H2O. Although the patient had no history of illness or immunization, she tested positive for severe acute respiratory syndrome-coronavirus-2 (SARS-CoV-2) IgG. Follow-up neuroimaging showed normal results, and the splenial lesion resolved within three weeks. SARS-CoV-2 infection may be related to reversible splenial lesion syndrome, and idiopathic intracranial hypertension is a rare occurrence.

Keywords: Child; COVID-19; pseudotumor cerebri; magnetic resonance imaging ÖZET Reversibl splenial lezyon sendromu, korpus kallozumun orta spleniumunda geri dönüşümlü bir lezyon ile karakterizedir. Enfeksiyonlar gibi farklı patolojilerle ilişkili olabilir. Çalışmada, baş dönmesi, ataksi ve görme bulanıklığı ile başvuran ve reversibl splenial lezyon gelişen bir kız çocuğu sunuyoruz. Lomber ponksiyon ile beyin omurilik sıvısı basıncının 39 cm H2O olduğu gösterildi. Enfeksiyon veya aşı öyküsü olmamasına rağmen şiddetli akut solunum sendromu-koronavirüs-2 [severe acute respiratory syndrome-coronavirus-2 (SARS-CoV-2)] IgG pozitifi. Üç hafta sonra kontrol nörogörüntülemede bulgular normaldi ve splenial lezyon görüntüsü gerilemişti. Reversibl splenial lezyon sendromu SARS-CoV-2 enfeksiyonu ile ilişkili olabilir ve idiyopatik intrakraniyal hipertansiyon birlikteliği nadir görülen bir durumdur.

Anahtar Kelimeler: Çocuk; COVID-19; psödotümör serebri; manyetik rezonans görüntüleme

Reversible splenial lesion syndrome (RESLES) is a type of lesion that occurs in the middle splenium of the corpus callosum (SCC), and it is characterized by reversible changes of unknown origin.<sup>1,2</sup> This condition has been associated with both infectious and non-infectious disorders, with reports of cases associated with viruses such as influenza, measles, mumps, rotavirus, adenovirus, Epstein-Barr virus, and herpesvirus 6.<sup>3,4</sup> The most common neurological symptoms of RESLES are encephalitis and encephalopathy while idiopathic intracranial hypertension is a rare complication.<sup>5-7</sup>

Although severe acute respiratory syndromecoronavirus-2 (SARS-CoV-2) primarily affects the lungs, there have been reports of various neurological complications, such as meningitis, ischemic stroke, encephalitis, and Guillain-Barré syndrome, in patients with coronavirus disease-2019 (COVID-19).<sup>8,9</sup> This article reports on the clinical symptoms of a girl with RESLES who was also found to be positive for SARS-CoV-2.

### CASE REPORT

A 14-year-old girl with no previous medical history presented with symptoms of dizziness, ataxia, and blurred vision for a duration of three weeks. Upon neurological examination, she exhibited confusion and ataxia, but her cranial nerves were intact, and she

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FIGURE 1: Brain magnetic resonance imaging of the patient. T2-weighted sagittal (A), axial (B) images of the patient show hyperintensity and restricted diffusion in the splenium of the corpus callosum. T2-weighted sagittal (C), axial (D) images of patient resolution of the lesion in the splenium of the corpus callosum.

had normal deep tendon reflexes and muscular tone. Pathologic reflexes were absent, but bilateral papilledema was discovered during an ophthalmologic examination. All standard laboratory tests came back normal, but a lumbar puncture showed that she had a cerebrospinal fluid pressure of 39 cm H<sub>2</sub>O. She had no prior history of SARS-CoV-2 infection or immunization, but a positive SARS-CoV-2 immunoglobulin (Ig) G test indicated that she had been infected with the virus. Polymerase chain reaction tests for SARS-CoV-2 on the patient's nasopharyngeal swab came back negative. Magnetic resonance imaging (MRI) of her brain showed hyperintensity on T2weighted images and restricted diffusion in the SCC (Figure 1). Treatment with acetazolamide and intravenous immunoglobulin (IVIG) was administered, and her symptoms subsided within a week. After three weeks, a follow-up MRI confirmed that the splenial lesion had resolved. The patient provided her consent for treatment and for the publication of her case.

The patient's parents have provided verbal and written consent for case report publication.

### DISCUSSION

We present a pediatric case of RESLES related to SARS-CoV-2 infection. The diagnosis was made after detection of anti-SARS-CoV-2 IgG. Interestingly, our case exhibited cerebellar ataxia as the initial symptom, which is an uncommon presentation of COVID-19. Hayashi et al. reported a COVID-19 patient with prior neurologic comorbidities who experienced a one-week remission of neurologic symptoms, while El Aoud et al. reported a one-week improvement in neurologic abnormalities in an adult patient with SARS-CoV-2 infection and resolution of the splenial lesion on MRI after one month, without any immunomodulatory therapy.<sup>10,11</sup> Our patient's symptoms improved within a week, and follow-up brain MRI three weeks later showed resolution of the splenial lesion. RESLES is a rare disorder associated with various pathological conditions and neurological symptoms, with a low incidence in adults. Persaud et al. reported the first case of RESLES with idiopathic intracranial hypertension, which was also rare in our case.<sup>12</sup> There is no established specific treatment for RESLES with neurological symptoms due to limited patient numbers. Antibiotics, acyclovir, anti-epileptic treatments, corticosteroids, and IVIG are the most commonly used therapies in such cases, according to the literature.<sup>13,14</sup> Our patient was treated with IVIG (2 g/kg) and acetazolamide. It is important to thoroughly analyze patients' MRI scans and employ repeated MRI scans for follow-up in order to ascertain the transient nature of such lesions. Further investigations on future cases could enhance our comprehension of RESLES.

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#### **Conflict of Interest**

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

#### Authorship Contributions

Idea/Concept: Kürşat Bora Çarman; Design: Kürşat Bora Çarman; Control/Supervision: Kürşat Bora Çarman; Data Collection and/or Processing: Dudu Şahin; Analysis and/or Interpretation: Arife Derda Yücel Şen; Literature Review: Arife Derda Yücel Şen; Writing the Article: Arife Derda Yücel Şen; Critical Review: Coşkun Yarar; References and Fundings: Kürşat Bora Çarman; Materials: Kürşat Bora Çarman.

### REFERENCES

- 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):230-3. [Crossref] [PubMed] [PMC]
- Kashiwagi M, Tanabe T, Oba C, Nomura S, Ashida A. Reversible splenial lesions during febrile illness with or without white matter lesions. Brain Dev. 2021;43(6):698-704. [Crossref] [PubMed]
- Bulakbasi N, Kocaoglu M, Tayfun C, Ucoz T. Transient splenial lesion of the corpus callosum in clinically mild influenza-associated encephalitis/ encephalopathy. AJNR Am J Neuroradiol. 2006;27(9):1983-6. [PubMed] [PMC]
- Karampatsas K, Spyridou C, Morrison IR, Tong CY, Prendergast AJ. Rotavirus-associated mild encephalopathy with a reversible splenial lesion (MERS)-case report and review of the literature. BMC Infect Dis. 2015;15:446. [Crossref] [PubMed] [PMC]
- Tsuji M, Yoshida T, Miyakoshi C, Haruta T. Is a reversible splenial lesion a sign of encephalopathy? Pediatr Neurol. 2009;41(2):143-5. [Crossref] [PubMed]
- 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] [PubMed]
- Degirmenci E, Degirmenci T, Cetin EN, Kıroğlu Y. Mild encephalitis/encephalopathy with a reversible splenial lesion (MERS) in a patient presenting with papilledema. Acta Neurol Belg. 2015;115(2):153-5. [Crossref] [PubMed]

- Mao L, Jin H, Wang M, Hu Y, Chen S, He Q, et al. Neurologic manifestations of hospitalized patients with coronavirus disease 2019 in Wuhan, China. JAMA Neurol. 2020;77(6):683-90. [Crossref] [PubMed] [PMC]
- Ahmad I, Rathore FA. Neurological manifestations and complications of COVID-19: a literature review. J Clin Neurosci. 2020;77:8-12. [Crossref] [PubMed] [PMC]
- Hayashi M, Sahashi Y, Baba Y, Okura H, Shimohata T. COVID-19-associated mild encephalitis/encephalopathy with a reversible splenial lesion. J Neurol Sci. 2020;415:116941. [Crossref] [PubMed] [PMC]
- El Aoud S, Sorial D, Selmaoui A, Menif I, Lazard M, Si Hocine M, et al. A first case of Mild Encephalitis with Reversible Splenial Lesion(MERS) as a presenting feature of SARS-CoV-2. Rev Neurol (Paris). 2021;177(1-2):139-41. Erratum in: Rev Neurol (Paris). 2021;177(8):1045. [Crossref] [PubMed] [PMC]
- Persaud A, Sakho F, Sandhu S, Krishnaiengar S. Reversible splenial lesion syndrome (RESLES) associated with Idiopathic Intracranial Hypertension: a rare case report and review of the literature. (P4.010). Neurology. 2018;90(15 Supplement). [Crossref]
- 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):1854-8. [Crossref] [PubMed]
- Fluss J, Ferey S, Menache-Starobinski C, Delavelle J, Van Bogaert P, Vargas MI. Mild influenza-associated encephalopathy/encephalitis with a reversible splenial lesion in a Caucasian child with additional cerebellar features. Eur J Paediatr Neurol. 2010;14(1):97-100. [Crossref] [PubMed]