

Vigabatrin Toxicity in a Patient with Infantile Spasms Treated with Concomitant Hormonal Therapy

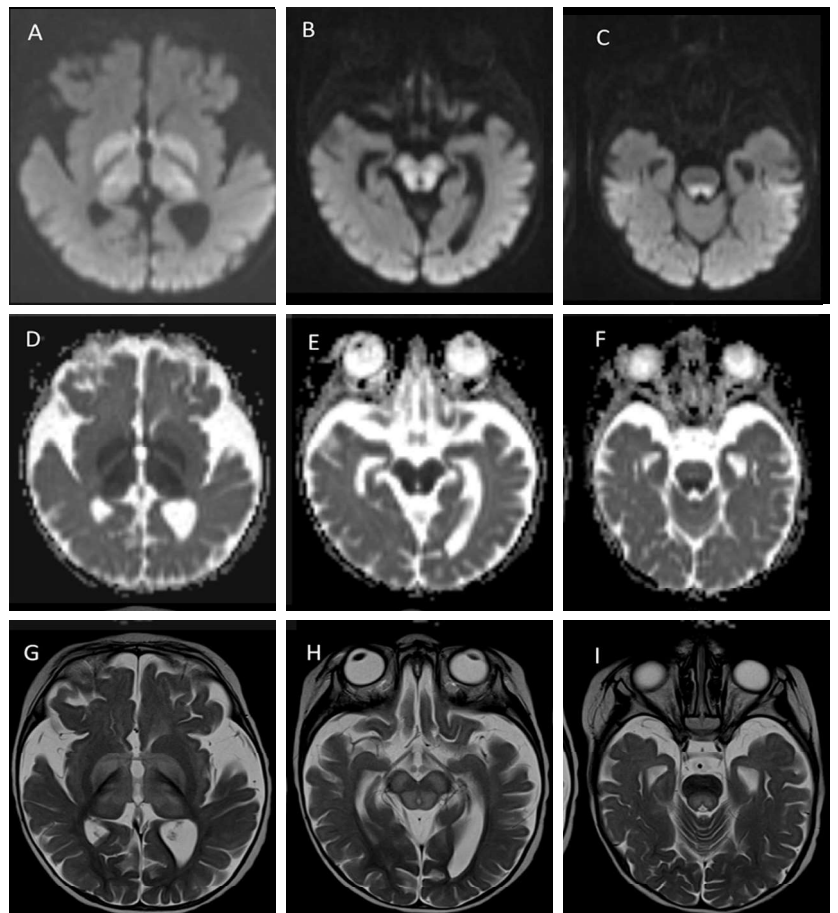
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An 8-month-old girl with infantile spasms, treated with adrenocorticotropic hormone and high-dose of vigabatrin (182 mg/kg/day), presented with a new movement disorder and marked encephalopathy. Magnetic resonance imaging (MRI) results showed restricted diffusion in the bilateral globus-pallidi, thalami, subthalamic regions, and dorsal brainstem [Figure 1], likely vigabatrin-related MRI signal changes, as sole treatment with adrenocorticotropic hormone [1] do not exhibit these changes [2]. Vigabatrin was discontinued due to the suspected toxicity [3], and clobazam was initiated with resolution of the clinical and MRI abnormalities [Figure 2]. It is important to distinguish reversible vigabatrin-associated brain MRI findings from other metabolic, toxic, ischemic, and infectious etiologies, especially as they appear to be dose-related and possibly more likely to be symptomatic with concomitant adrenocorticotropic hormone therapy [4].

Figure 1. Brain magnetic resonance imaging (MRI) findings 8 weeks after initiation of therapy with vigabatrin and adrenocorticotropic hormone. Axial diffusion-weighted images [A] [B] [C], ADC maps [D] [E] [F], and axial T2-weighted images [G] [H] [I] of the brain at 8 months of age show restricted diffusion and abnormal T2 hyperintense signal in bilateral globus-pallidi, thalami, subthalamic regions, and dorsal brainstem



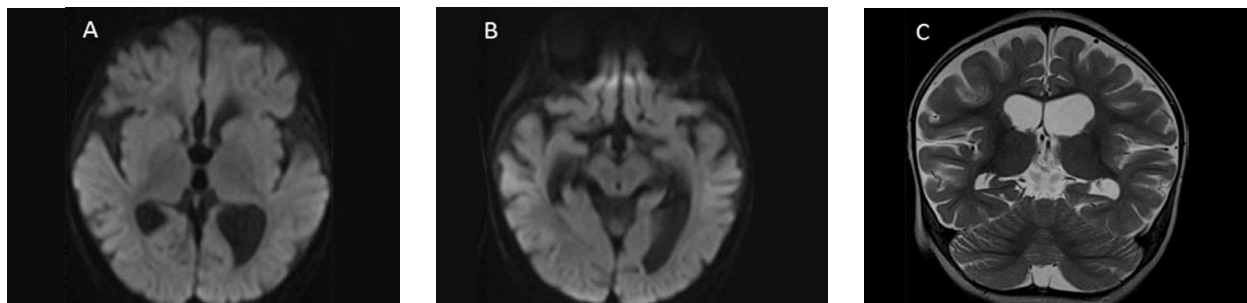
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References

1. Waternberg N. The use of steroids in epilepsy: time for a reappraisal? *IMAJ* 2010; 12 (3): 176-7.
2. Dracopoulos A, Widjaja E, Raybaud C, Westall CA, Snead OC 3rd. Vigabatrin-associated reversible MRI signal changes in patients with infantile spasms. *Epilepsia* 2010; 51: 1297-304.
3. Dill P, Datta AN, Weber P, Schneider J. Are vigabatrin induced T2 hyperintensities in cranial MRI associated with acute encephalopathy and extrapyramidal symptoms? *Eur J Paediatr Neurol* 2013; 17: 311-15.
4. Hussain SA, Tsao J, Li M, et al. Risk of vigabatrin-associated brain abnormalities on MRI in the treatment of infantile spasms is dose-dependent. *Epilepsia* 2017; 58: 674-82.

Figure 2. Follow-up brain magnetic resonance imaging (MRI) 10 months after discontinuation of vigabatrin therapy. Axial diffusion weighted images [A] [B] and coronal T2-weighted image [C] of the brain at 18 months of age show resolution of the previously seen abnormalities in the deep gray nuclei and brainstem



Capsule

A public health catch-22

In 2016, the serotype 2 component of the oral poliovirus vaccine given to children was withdrawn. This measure was taken to prevent vaccine-associated disease outbreaks caused by mutation in the live attenuated vaccine. Children around the world now have poor immunity to serotype 2 poliovirus because the inactivated vaccine is far less effective and a new oral vaccine is not yet ready. Using a statistical model, **Macklin** and co-authors discovered that most current outbreaks of

polio in several countries across Asia and sub-Saharan Africa are likely associated with the serotype 2 vaccine strain. To block transmission when poliovirus outbreaks occur requires deployment of the only tool in the box: the existing live attenuated serotype 2 oral vaccine, which increases the risk of vaccine-derived disease.

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Capsule

Clinical characteristics and results of semen tests among men with coronavirus disease 2019

The virus responsible for COVID-19, severe acute respiratory syndrome-coronavirus 2 (SARS-CoV-2) has been detected in stool, gastrointestinal tract, saliva, and urine samples. However, little is known about SARS-CoV-2 in semen. Of these 38 participants who provided a semen specimen, 23 participants (60.5%) had achieved clinical recovery and 15 participants (39.5%) were at the acute stage of infection. Results of semen testing found that 6 patients (15.8%) had results positive for SARS-CoV-2, including 4 of 15 patients (26.7%) who were at the acute stage of infection and 2 of 23 patients (8.7%) who were recovering, which is particularly noteworthy. There was no significant difference between negative and positive test results for patients by age, urogenital disease history, days since onset, days since hospitalization, or days since clinical recovery.

The authors found that SARS-CoV-2 can be present in the semen of patients with COVID-19, and SARS-CoV-2 may still be detected in the semen of recovering patients. Due to the imperfect blood-testes/deferens/epididymis barriers, SARS-CoV-2 might be seeded to the male reproductive tract, especially in the presence of systemic local inflammation. Even if the virus cannot replicate in the male reproductive system, it may persist, possibly resulting from the privileged immunity of testes. This finding does not prove that SARS-CoV-2 can be transmitted sexually. Abstinence or condom use might be considered as preventive means for these patients. In addition, it is worth noting that there is a need for studies monitoring fetal development.

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