Journal of Neuroradiology 46 (2019) 222–223

Available online at ScienceDirect www.sciencedirect.com

Elsevier Masson France EM|consulte www.em-consulte.com

Correspondence

Cytotoxic lesion of the corpus callosum exclusively at the genu in a case of callosal hypogenesis

Introduction

Cytotoxic lesions of the corpus callosum (CLOCCs) are secondary lesions associated with many entities, including infection, drug therapy, malignancy, metabolic disorders and others [1,2]. Although CLOCCs extending to the entire corpus callosum (CC), and occasional bilateral white matter and basal ganglia involvement were previously observed [3,4], isolated lesions in the genu during the acute stage have not been reported. Here we first report transient CLOCC showing an isolated genu lesion, in a patient with interhemispheric cyst and callosal hypogenesis.

Case report

A previously healthy 4-year-old male patient was admitted due to impaired consciousness and seizures, following prodromal symptoms of fever and upper respiratory tract infection. Neurological examination was unremarkable except for decreased consciousness due to postictal state lasting more than 24 hours. Except for mild hyponatremia (Na = 132 mEq/L), laboratory tests were negative for metabolic disorders. Electroencephalography showed bilateral slow waves with right predominance. Brain MRI revealed an interhemispheric cyst with callosal hypogenesis (Fig. 1A–D). The body of the CC was almost absent, and a hypoplastic splenium was observed (Fig. 1B–C). Diffusion-weighted images (DWI) showed hyperintense signal in the genu with a reduced apparent diffusion coefficient (ADC) (Fig. 1F–G). Other MRI sequences (T1-MPRAGE, T2-FLAIR, SWI) failed to detect other abnormalities including cortical dysplasia. Cerebrospinal fluid analysis was not conducted because his level of consciousness improved gradually without notable meningeal symptoms, and he totally recovered on day 2. Nasopharyngeal and blood culture, as well as antigen diagnostic tests for influenza virus and adenovirus were negative. Follow-up MRI performed on day 7 showed complete resolution of signal intensities (Fig. 1H–I), and an electroencephalogram performed on day 30 was normal. A final diagnosis of CLOCC with an isolated genu lesion was made based on the clinical and radiological features. Antiepileptic drug was not administered, and no neurological complications were observed at the last follow-up.

Discussion

CLOCCs are observed in various entities, also known as mild encephalopathy with reversible splenial lesions (MERS), or reversible splenial lesion syndrome (RESLES) in different regions [1,2]. The pathophysiological mechanism of CLOCCs is currently not completely understood. Various unspecific pathogens have been reported as CLOCCs-assosiated infectious agents [5]. Genetic factors have also been suggested. A causal association for MYRF was recently reported in siblings showing CLOCCs, but such association was not observed in isolated cases [3]. Vulnerability to cytokinopathy was postulated as possible reason for the regional specificity seen in the splenium of the corpus callosum (SCC) [2].

The CC, the largest white matter tract in the human brain, connects the cerebral hemispheres, and provides interhemispheric integration and information transfer [6]. Research into the normal postnatal development of the CC showed that by 12 months, the thickness of splenium is slightly higher than that of the genu [7]. A recent report using older subjects (5–18 years) showed that maximum growth of the CC was detected in the splenium [6]. Taken together, the SCC is morphologically thickest in children after 12 months of age, a result of the myelination process concurrent with callosal maturation [6,7].

In cases with CLOCCs without major callosal structural abnormalities, the SCC is consistently reported as the prime lesion site [1,2]. In the present case, the lesion was located at the genu (9.4 mm), which was thicker than the hypoplastic splenium (6.7 mm) due to callosal hypogenesis. Compared to other types of acute encephalopathy (median age, 1–2 years), the median age at onset of MERS patients was previously reported to be 5 years [4]. This suggests that CLOCCs usually occur after the maturation of CC with sufficient myelin volume. Our finding suggests that the regional specificity of the reversible lesions seen in CLOCCs might be determined by the myelin volume. Further research employing methods of myelin volume quantitative analysis is required to confirm this hypothesis.

Funding

All authors certify they have no financial interest to disclose.

Disclosure of interest

The authors declare that they have no competing interest.

https://doi.org/10.1016/j.neurad.2018.11.001

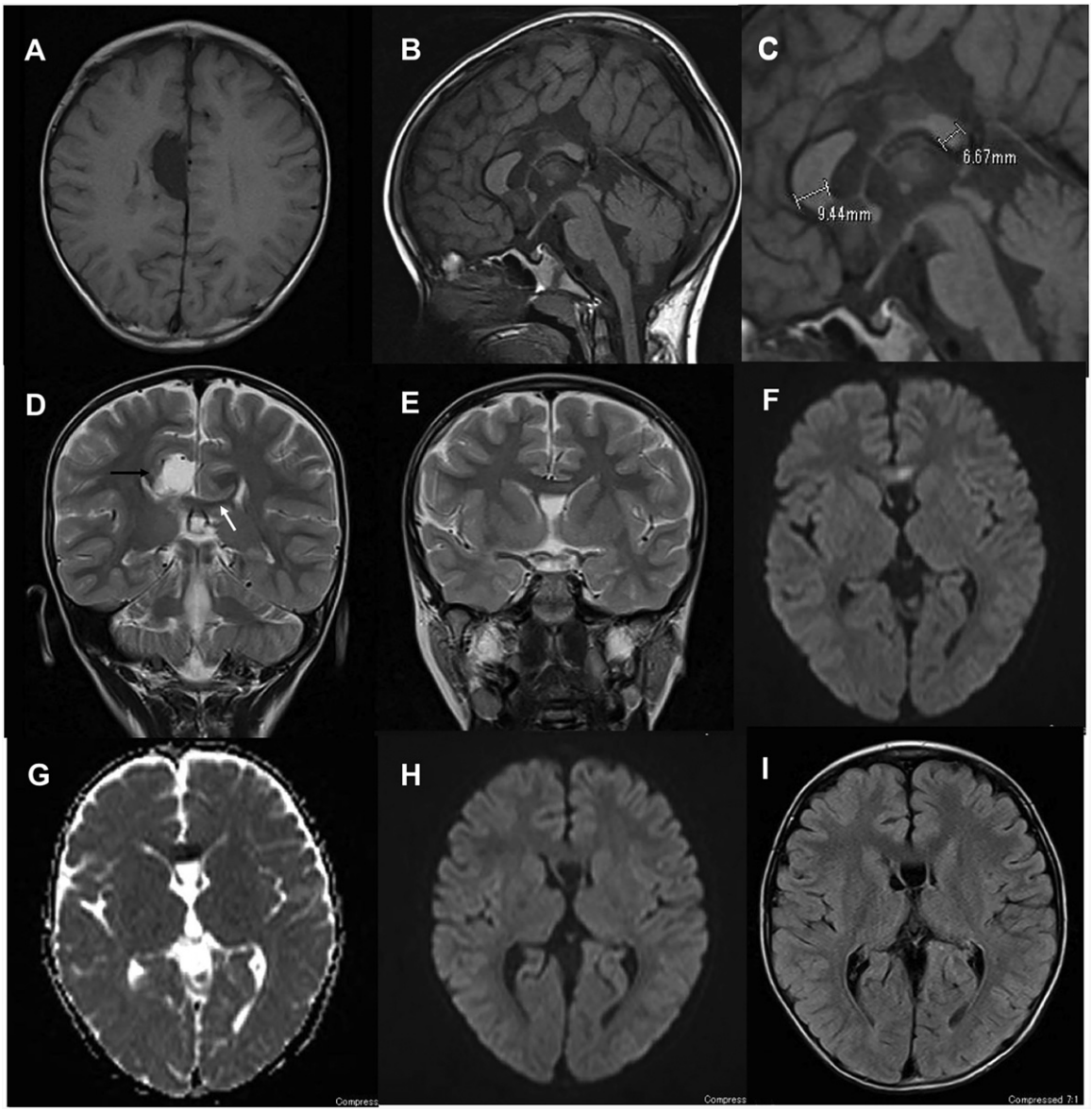

Fig. 1. Brain MRI findings. Axial T1-weighted (A) scan shows right interhemispheric cyst adjacent to the falx cerebri. Sagittal T1-weighted scan (B) shows callosal hypogenesis with a nearly absent razor-thin body and hypoplastic splenium, compared to a relatively normally sized genu. Thickness of the genu and splenium were measured as shown in the enlarged figure (C). Coronal T2-weighted scans at the splenium level (D) and genu level (E) show thinning of the axonal fiber of the splenium (white arrow). The interhemispheric cyst is indicated by the black arrow. Significant signal hyperintensity on DWI (F) and hypointensity on ADC (G) was observed in the genu at admission. The hyperintensity signal on DWI of the isolated genu lesion disappeared on day 7 (H). No lesion was noted on corresponding axial T2-FLAIR scan (I).

References

[1] 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;63:1854–8.
[2] Starkey J, Kobayashi N, Numaguchi Y, Moritani T. Cytotoxic lesions of the corpus callosum that show restricted diffusion: mechanisms, causes, and manifestations. Radiographics 2017;37:562–76.
[3] Kurahashi H, Azuma Y, Masuda A, Okuno A, Nakahara E, Imamura T, et al. MYRF is associated with encephalopathy with reversible myelin vacuolization. Ann Neurol 2018;83:99–106.
[4] Renard D, Taieb G, Briere C, Bengler C, Castelnovo G. Mild encephalitis / encephalopathy with a reversible splenial, white matter, putaminal and thalamic lesions following anti-Yo rhomboencephalitis. Acta Neurol Belg 2012;112:405–7.
[5] Hoshino A, Saitoh M, Oka A, Okumura A, Kubota M, Saito Y, et al. Epidemiology of acute encephalopathy in Japan, with emphasis on the association of viruses and syndromes. Brain Dev 2012;34:337–43.
[6] Luders E, Thompson PM, Toga AW. The development of the corpus callosum in the healthy human brain. J Neurosci 2010;30:10985–90.
[7] Barkovich AJ, Kjos BO. Normal postnatal development of the corpus callosum as demonstrated by MR imaging. AJNR Am J Neuroradiol 1988;9:487–91.

Moeri Tsuji
Pin Fee Chong*
Fumiya Yamashita
Kenichi Maeda
Ryutaro Kira
Department of Pediatric Neurology, Fukuoka Children's Hospital, Fukuoka, Japan

* Corresponding author at: 5-1-1, Kashiiteriha, Higashi-ku, Fukuoka 8130017, Japan.
E-mail address: chong.p.f@fcho.jp (P.F. Chong)

Available online 27 November 2018