

Case report

Cerebral embolism accompanied by remarkable diffusion-weighted imaging reversal in a 10 month-old infant with congenital heart disease

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1. Introduction

Cyanotic congenital heart disease is one of the main causes of juvenile ischemic stroke and sometimes accompanies chronic hypoxia. Herein, we describe a patient who had a cerebral embolism and showed diffusion-weighted imaging (DWI) reversal.

2. Case report

A 7 month-old boy underwent the Glenn procedure for right ventricular hypoplasia and multiple large ventricular septal defects. To shrink the defects before the Fontan procedure, an intracardiac blind pouch at the pulmonary artery trunk was formulated, and warfarin was started for thrombosis prevention. Three months later (at 10 months of age), he suddenly developed left hemiparesis and visited our emergency room. Neurological findings revealed slight left hemiparesis and no apparent spatial neglect. Laboratory findings showed hypoxia (pO₂ 50 mmHg) and mild prolonged Prothrombin Time and International Normalized Ratio (PT-INR) (1.27). Other coagulation markers were normal. DWI performed 10 h after symptom onset showed acute cerebral infarcts in the left middle cerebral artery (MCA) territory, and the DWI-Alberta Stroke Program Early Computed Tomography Score (DWI-ASPECTS) was 6. Core volume was 13 ml with automated software (RAPID, iSchemaView, Menlo Park, CA, version 4.7) and was smaller than the visually identified DWI lesion (Fig. 1A). Because magnetic resonance angiography and carotid ultrasonography revealed no occlusion or stenosis in the main arteries, early spontaneous recanalization after embolization was suspected. No atrial fibrillation or deep vein thrombosis was present, nor did we observe an apparent thrombus at the blind pouch at the pulmonary artery trunk. We diagnosed this case with cerebral embolism due to a right-to-left shunt associated with congenital heart disease during subtle anticoagulation therapy. His left hemiparesis gradually improved and completely disappeared during admission. Follow-up magnetic resonance imaging (MRI) at 7 days revealed no DWI-positive lesion.

3. Discussion

We report a case of ischemic stroke in an infant with cyanotic congenital heart disease accompanied by chronic hypoxia who had a remarkable DWI reversal change. DWI reversal was previously reported in adult cases; however, this is the first case that confirmed DWI reversal in an infant.

DWI reversal is observed in lesions with a less severe initial apparent diffusion coefficient (ADC) decrease than persistent lesions [1]. Although ADC is a biomarker of cytotoxic injury, ADC values above 80% of baseline are associated with normalization following reperfusion [2]. Therefore, a mismatch seen in automated core estimation and visually identified DWI lesions may reflect a lesion with less of an ADC decline, and this mismatch may predict DWI reversal. In addition, DWI reversal was observed in a large lesion in this case. Early recanalization [3] and chronic cerebral hypoxia caused by unrepaired cyanotic congenital heart disease after the Glenn procedure, which may have induced hypoxia preconditioning and ischemic tolerance [4,5], may contribute to large DWI reversal.

DWI-ASPECTS and the core volume show a weak correlation when DWI-ASPECTS is under 6, which is approximately > 33 ml in the adult ischemic stroke population [6]. Blurry DWI lesions are sometimes biased by ASPECT scoring, and on the other hand, are underestimated by automated software. In this case, however, a sufficient mismatch was present between DWI-ASPECTS and the core volume. The small volume of the infant brain may affect this mismatch. The interpretation of ASPECTS has not been well discussed in this population. Therefore, clinicians should carefully assess DWI-ASPECTS and the core volume, especially in infants with ischemic stroke.

In conclusion, a mismatch between automated core estimation and a visually identified DWI lesion may be essential for predicting DWI reversal to estimate the ADC changes behind the thresholds.

Ethical standards

The manuscript contains patient data.

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<https://doi.org/10.1016/j.ensci.2020.100260>

Received 18 May 2020; Received in revised form 30 June 2020; Accepted 29 July 2020

Available online 12 August 2020

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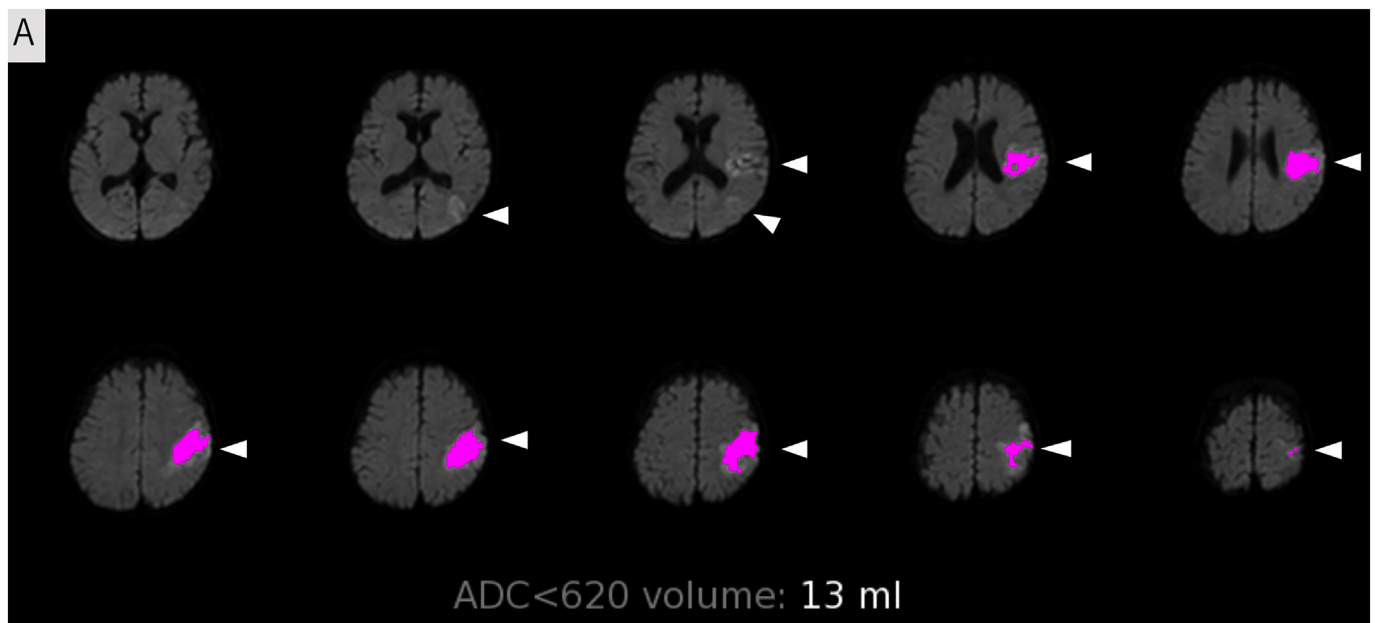


Fig. 1. A: DWI demonstrating a high intensity in the left MCA territory (indicated by arrowheads). Although a broad DWI hyperintense lesion was present in the left MCA territory, the core volume calculated by RAPID showed an ADC declined lesion of 13 ml (indicated by the pink area), and a mismatch between the visually identified and calculated DWI lesion was observed. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Declaration of Competing Interest

The authors declare that they have no conflict of interest.

Acknowledgements

We wish to thank the patient and his family for their cooperation regarding this report. We obtained written informed consent for this publication.

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