Neurology Asia 2017; 22(2) : 149 – 154

CASE REPORTS

Absence of hemispatial neglect following right hemispherectomy in a 7-year-old girl with neonatal stroke

*[1]Jiqing Qiu *PhD*, *[2]Yu Cui *MD*, [3]Lichao Sun *MD*, [1]Bin Qi*PhD*, [1]Zhanpeng Zhu *PhD*
*JQ Qiu and Y Cui contributed equally to this work

Departments of [1]Neurosurgery, [2]Otolaryngology and [3]Emergency Medicine, The First Hospital of Jilin University, Changchun, Jilin, China

Abstract

Neonatal stroke leads to cognitive deficits that may include hemispatial neglect. Hemispatial neglect is a syndrome after stroke that patients fail to be aware of stimuli on the side of space and body opposite a brain lesion. We report here a 7-year-old girl who suffered neonatal right brain stroke and underwent right hemispherectomy due to refractory epilepsy. Post-surgical observation of the child's behavior and tests did not show any signs of hemispatial neglect. We concluded the spatial attention function of the child with neonatal stroke might be transferred to the contralateral side during early childhood.

Key words: Epilepsy; hemispatial neglect; hemispherectomy; neonatal stroke; child.

INTRODUCTION

Approximately one in every 4,000 newborns suffer from neonatal stroke, with a high mortalityrate.[1-3] The survivors may develop varying degrees of hemiparesis and low intelligence quotient (IQ). Children with neonatal stroke, especially children with hemiplegia, have a high risk of developing cognitive impairments.[4-7] Cognitive deficits in stroke patients include disorders of spatial awareness, known as hemispatial neglect.[8,9] Hemispatial neglect refers to the impairment or loss in ability to perceive and process visual, auditory, tactile, or olfactory sensory stimuli presented in the hemispace contralateral to a lesion of one cerebral hemisphere.[10]

Some children with neonatal stroke develop seizures[11] and may consequently undergo hemispherectomy during childhood.[12]Current studies on neonatal stroke focus mainly on the status of speech/motor and sensory function. Little is known about development of hemispatial neglect. Since the brain shows greater plasticity in the early childhood, we would like to report the effect of the neonatal stroke on spatial attention of a child post-hemispherectomy.

CASE REPORT

The patient was a 7-year-old right-handed girl. She was delivered with normal body weight. Gestation and delivery were uneventful. There was no family history of epilepsy. She had coma a few days after birth and was diagnosed with neonatal stroke (Figure 1A-D). She woke up 7 days later, with left-sided hemiparesis.

At two and half years old, she developed simple partial seizures, with a frequency of 5-6 times per day. She came to our Hospital, and was treated with topiramate. At age of 4 years, her seizure type changed to that of complex partial seizures. Lamotrigine was added to topiramate. Most of the seizures were nocturnal during sleep. The seizures may be provoked by sudden noise, such as the sound of a door closing. The patient was compliant with the antiepileptic drugs.

When the patient was seen at age 7 years, she was taking topiramate (100 mg bd) and lamotrigine (60 mg bd). Despite the medications, she had 7-8 seizures per day. Brain MR imaging revealed a hyperintense and atrophied right hemisphere, particularly the temporo-parieto-occipital regions (Figure 2A, B). CT angiography showed that the branches of the right middle cerebral artery were

Address correspondence to: Zhanpeng Zhu, Department of Neurosurgery, The First Hospital of Jilin University, Changchun 130021, China. Email: 282324491@qq.com

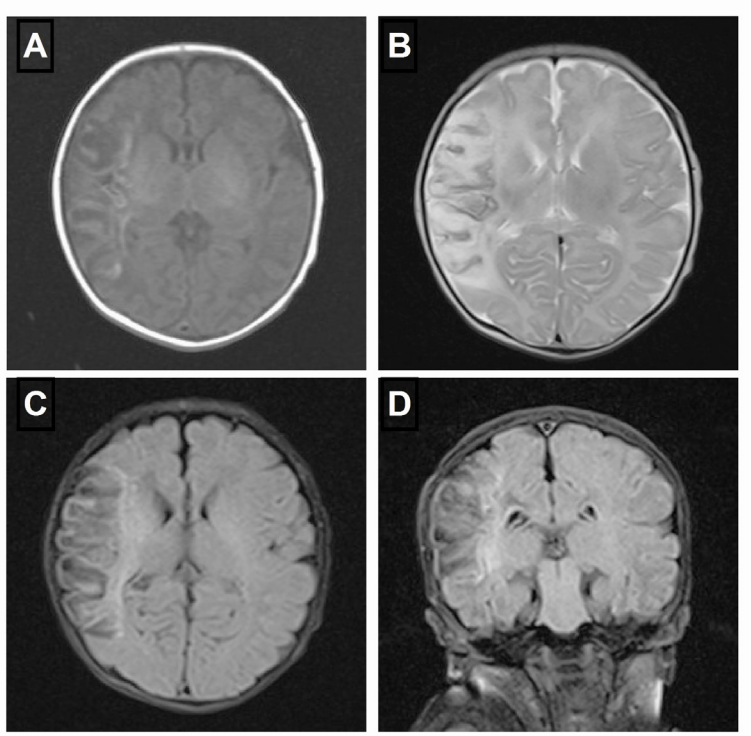


Figure 1. Neuroimaging when patient was in coma, MRI axial T_1 (A), axial T_2 (B), axial T_2 flair (C) and coronal T_2 flair (D) showing there was a ischemic stroke in the right hemisphere.

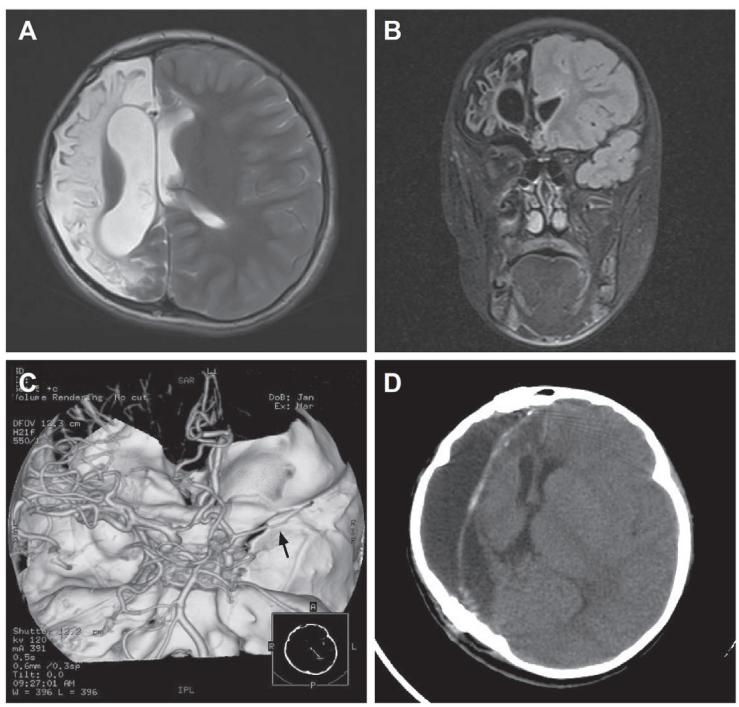


Figure 2. (A), (B) Presurgical neuroimaging: MRI axial $T_2$and coronal T1 showing the extent of the lesion in the right hemisphere. The skull was asymmetric and a large porencephalic cyst was present. (C) Presurgical neuroimaging (head CT angiography): the branches of the right middle cerebral artery were hypoplastic or absent (arrow). (D) Post-hemispherectomy neuroimaging: CT head scan showing the right anatomical hemispherectomy.

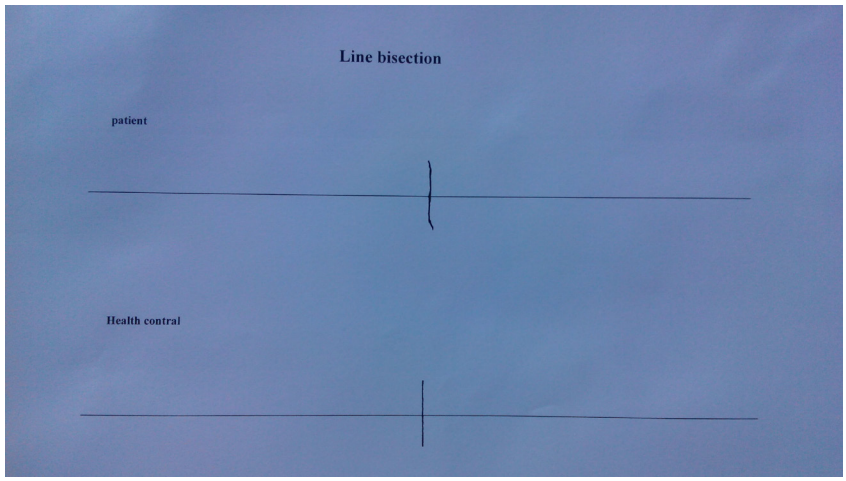
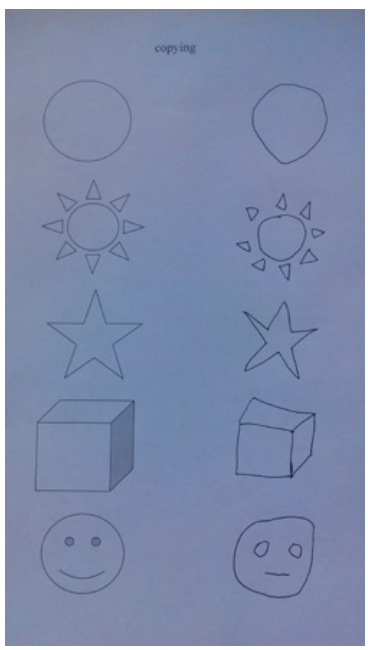


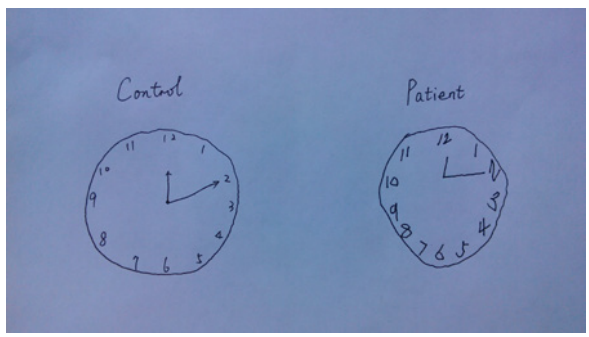
Figure 3. The patient's normal line bisection before surgery showed that the patient did not have hemianopia and unilateral neglect.

slender (Figure 2C). The normal line bisection test done before surgery was shown in Figure 3.

Her laboratory investigations were normal. Due to the intractable epilepsy, she underwent an anatomical right hemispherectomy. The cortex of the right hemisphere showed some atrophy and was removed, leaving the thalamus and basal ganglia intact (Figure 2D). After surgery, all pre-surgical function, including sensory and motor functions were retained. Based on the Gaze Orientation Scale that assesses spontaneous gaze and head orientation for neglect, no deviation of gaze and head was observed pre- or post-surgery. After the surgery, she demonstrated no signs of hemispatial neglect. Specifically, when asked to identify ten objects around the room, she could name the items on both the left and right sides. She could also grasp toys around her body. When asked to comb her hair or makeup her face, she could groom herself on both sides. Three months after the surgery, she was given the drawing tests (Figure 4) and cancellation tests (Figure 5) that examines unilateral spatial neglect in children, no abnormality was observed.

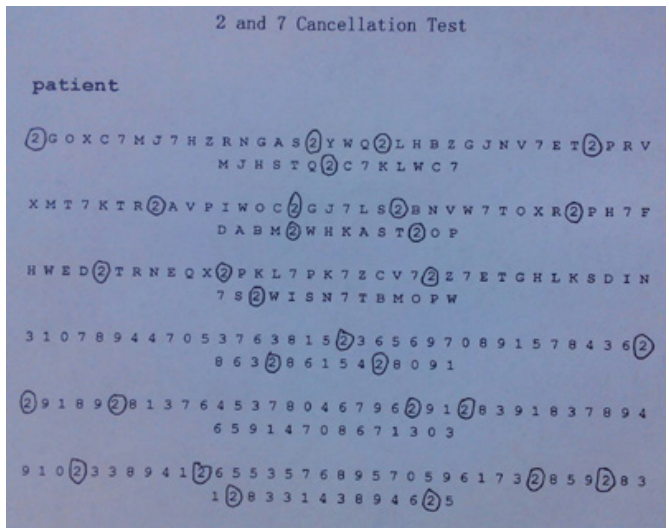


(A)

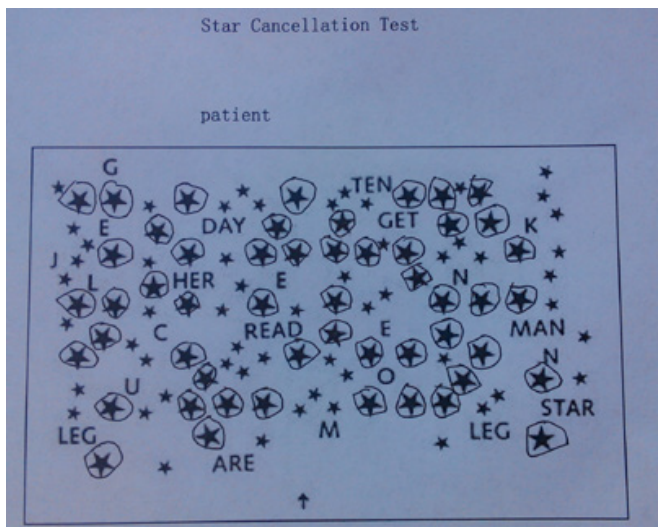


(B)

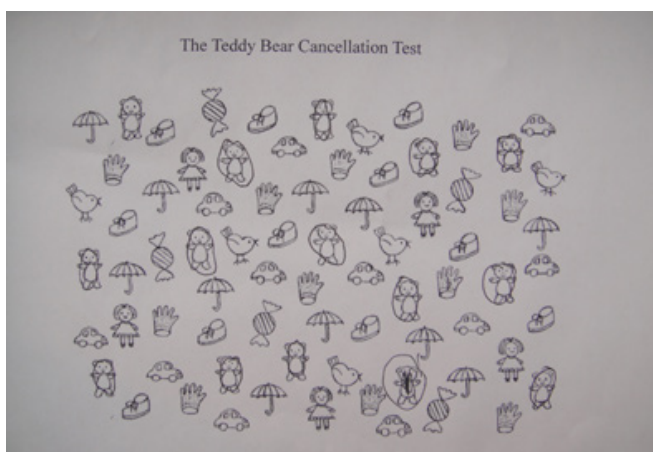
Figure 4. (A), (B). The patient's normal clock drawing test performed three months after right hemispherectomy



(A)



(B)



(C)

Figure 5 (A), (B), (C). The patient's normal cancellation tests performed three months post-hemispherectomy

DISCUSSION

Neonatal stroke is an important cause of chronic neurological morbidity in children. It has been shown that 57% of the infants with neonatal stroke were neurologically or cognitively abnormal.[3] Some children with neonatal stroke develop hemiparesis or seizures during childhood. Persistent seizures during childhood could slow down cognitive development.[13,14] Previous studies have indicated that children with a longer period of active epilepsy showed lower IQs than children with a shorter seizure history.[15]

Hemispherectomy may be a useful treatment modality for some congenital or acquired medical intractable epilepsy.[16,17] It involves complete removal (anatomical hemispherectomy) or partial resection and disconnection (functional hemispherectomy) of the cerebral hemisphere. We performed anatomical hemispherectomy for our patient because of medical intractable epilepsy.

The child who undergo hemispherectomy for treatment is missing the entire cortex on one side after surgery[18], all functions are then dependent on a single hemisphere. Lesions in the right hemisphere often lead to hemispatial neglect since the right hemisphere is predominantly responsible for spatial attention[8,9,19,20], with lesions involving right inferior parietal and adjacent temporal lobe being associated with spatial neglect.[21] Thus, the right anatomical hemispherectomy provides an opportunity to examine whether spatial attention function has been transferred to the contralateral side in the neonatal stroke child before the surgery.

The clock drawing test[22] has been suggested as a valuable tool for screening the hemispatial neglect. It has been reported that the clock drawing test can identify spatial neglect in children with early hemispheric damage.[23] Our patient could draw a circle clock and do not show unilateral spatial neglect. She also did well in other tests (cancellation tests and line bisection test) for hemispatial neglect. Line bisection could also detect hemianopia, thus differentiating it from hemispatial neglect.[24] As our patient did not show hemispatial neglect after right hemispherectomy, this suggested that at least part of the spatial attention function was transferred to the left hemisphere before surgery.

Our patient's spatial attention might have transferred to the contralateral side before 7-year-old. This is the same to previous studies.[25] Any cognitive reorganizing she has had may be a result of the neonatal stroke she suffered a few days after birth. Yet, there are case studies of pediatric patients with stroke whose neglect symptoms subsequently resolve.[26] Previous studies have suggested that long-term repeated seizures may lead to changes in brain function.[27] So seizures may also have contributed to the transferring of brain function to the contralateral side.

ACKNOWLEDGEMENTS

This study was approved by the ethics committee of The First Hospital of Jilin University. Informed parental consent was obtained in this case. The corresponding authors confirm that we have received a signed release form from the patient parents authorizing the publication of her material.

DISCLOSURE

Funding support: None

Conflict of interest: None

REFERENCES

1. Lee J, Croen LA, Lindan C, *et al*. Predictors of outcome in perinatal arterial stroke: a population-based study. *Ann Neurol* 2005;58:303-8.
2. Nelson KB, Lynch JK. Stroke in newborn infants. *Lancet Neurol* 2004; 3:150-8.
3. Lynch JK, Nelson KB. Epidemiology of perinatal stroke. *Curr Opin Pediatr* 2001; 13:499-505.
4. Westmacott R, MacGregor D, Askalan R, deVeber G. Late emergence of cognitive deficits after unilateral neonatal stroke. *Stroke* 2009;40:2012-9.
5. Aden U. Neonatal stroke is not a harmless condition. *Stroke* 2009; 40:1948-9.
6. Grunt S, Mazenauer L, Buerki SE, *et al*. Incidence and outcomes of symptomatic neonatal arterial ischemic stroke. *Pediatrics* 2015;135:e1220-8.
7. Chabrier S, Husson B, Dinomais M, Landrieu P, Nguyen The Tich S. New insights (and new interrogations) in perinatal arterial ischemic stroke. *Thromb Res* 2011;127:13-22.
8. Malhotra P, Coulthard E, Husain M. Hemispatial neglect, balance and eye-movement control. *Curr Opin Neurol* 2006; 19:14-20.
9. Adair JC, Barrett AM. Spatial neglect: clinical and neuroscience review: a wealth of information on the poverty of spatial attention. *Ann N Y Acad Sci* 2008;1142:21-43.
10. Kerkhoff G. Spatial hemineglect in humans. Prog Neurobiol 2001; 63:1-27.
11. Lehman LL, Rivkin MJ. Perinatal arterial ischemic stroke: presentation, risk factors, evaluation, and outcome. *Pediatr Neurol* 2014; 51:760-8.
12. Griessenauer CJ, Salam S, Hendrix P, *et al*. Hemispherectomy for treatment of refractory epilepsy in the pediatric age group: a systematic review. *J Neurosurg Pediatr* 2015;15:34-44.
13. Hirsch E, de Saint-Martin A, Arzimanoglou A. New insights into the clinical management of partial epilepsies. *Epilepsia* 2000; 41(Suppl 5):S13-7.

14. Oguni H, Mukahira K, Tanaka T, *et al*. Surgical indication for refractory childhood epilepsy. *Epilepsia* 2000; 41 (Suppl 9):21-5.
15. Robinson S, Park TS, Blackburn LB, Bourgeois BF, Arnold ST, Dodson WE. Transparahippocampal selective amygdalohippocampectomy in children and adolescents: efficacy of the procedure and cognitive morbidity in patients. *J Neurosurg* 2000; 93:402-9.
16. Anan M, Kamida T, Abe E, *et al*. A hemispherotomy for intractable startle epilepsy characterized by infantile hemiplegia and drop attacks. *J Clin Neurosci* 2009; 16:1652-5.
17. Liegeois F, Morgan AT, Stewart LH, Helen Cross J, Vogel AP, Vargha-Khadem F. Speech and oral motor profile after childhood hemispherectomy. *Brain Lang* 2010; 114:126-34.
18. Bahuleyan B, Robinson S, Nair AR, Sivanandapanicker JL, Cohen AR. Anatomic hemispherectomy: historical perspective. *World Neurosurg* 2013;80:396-8.
19. Heilman KM, Valenstein E, Watson RT. Neglect and related disorders. *Semin Neurol* 2000; 20:463-70.
20. Corbetta M. Hemispatial neglect: clinic, pathogenesis, and treatment. *Semin Neurol* 2014; 34:514-23.
21. Karnath HO, Niemeier M, Dichgans J. Space exploration in neglect. *Brain* 1998; 121 (Pt 12):2357-67.
22. Agrell B, Dehlin O. The clock-drawing test. 1998. *Age Ageing* 2012; 41(Suppl 3):iii41-5.
23. Yousefian O, Ballantyne AO, Doo A, Trauner DA. Clock drawing in children with perinatal stroke. *Pediatr Neurol* 2015; 52:592-8.
24. Lanyon LJ, Barton JJ. Visual search and line bisection in hemianopia: computational modelling of cortical compensatory mechanisms and comparison with hemineglect. *Plos one* 2013; 8(2):e54919.
25. Marsh EB, Newhart M, Kleinman JT, *et al*. Hemispherectomy sustained before adulthood does not cause persistent hemispatial neglect. *Cortex* 2009; 45:677-85.
26. Kleinman JT, Gailloud P, Jordan LC. Recovery from spatial neglect and hemiplegia in a child despite a large anterior circulation stroke and Wallerian degeneration. *J Child Neurol* 2010; 25:500-3.
27. Jokeit H, Ebner A. Effects of chronic epilepsy on intellectual functions. *Prog Brain Res* 2002; 135:455-63.