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● *Original Contribution*

ULTRASONOGRAPHIC CHANGES AFTER INDIRECT REVASCULARIZATION SURGERY IN PEDIATRIC PATIENTS WITH MOYAMOYA DISEASE

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Abstract—The marked cerebral hypoperfusion of moyamoya disease (MMD) can be treated with encephaloduroarteriosynangiosis (EDAS), an indirect revascularization surgery. Collateral establishment after the surgery is a gradual process; thus, easy access to serial assessment is of great importance. We prospectively recruited 15 pediatric moyamoya patients who underwent EDAS surgeries on a total of 19 hemispheres. Ultrasonography of extracranial and intracranial arteries was performed pre-operatively and post-operatively at 1, 3 and 6 mo. Among the extracranial arteries, the superficial temporal artery had the most pronounced increase in flow velocity and decrease in flow resistance from 1 mo post-surgery ($p < 0.01$). Among the large intracranial arteries, a significant increase in peak systolic velocity was observed in the anterior cerebral artery from 3 mo post-surgery ($p < 0.05$). These findings indicate significant hemodynamic changes on ultrasonography in pediatric moyamoya patients after indirect revascularization surgery. (E-mail: jsjeng@ntu.edu.tw) © 2016 World Federation for Ultrasound in Medicine & Biology.

Key Words: **Moyamoya disease, Pediatric stroke, Encephaloduroarteriosynangiosis, Indirect revascularization, Ultrasonography.**

INTRODUCTION

Moyamoya disease (MMD) is characterized by idiopathic chronic progressive stenosis or occlusion of bilateral distal internal carotid arteries (ICAs) and their major branches (Suzuki and Takaku 1969). The marked cerebral hypoperfusion in moyamoya patients usually presents as ischemic stroke (IS) or transient ischemic attack (TIA) (Scott and Smith 2009). The incidence and prevalence of MMD are higher in Asian populations (Baba et al. 2008; Chen et al. 2014; Duan et al. 2012; Han et al. 2000); nonetheless, this disease is also a major cause of pediatric stroke in Western countries (Currie et al. 2011). The ischemic events associated with MMD can be reduced by extracranial–intracranial (EC-IC) bypass surgery, including direct, indirect and combined surgeries (Bao et al. 2015).

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Moyamoya disease has a progressive course (Suzuki and Takaku 1969), which makes follow-up mandatory for this disease. Cerebral angiography is the gold standard tool for diagnosis and follow-up for MMD; however, the risks of permanent neurologic deficit and radiation exposure are ongoing safety concerns (Perren et al. 2005). Ultrasound is another non-invasive tool for the evaluation of hemodynamics; however, for MMD it is reported only for baseline status (Lee et al. 2004; Ruan et al. 2006) or for evaluations conducted after direct bypass surgery (Kraemer et al. 2012; Wu et al. 2011) but not for indirect revascularization surgery. The baseline characteristics revealed by ultrasound include an increase in resistance and a decrease in flow velocity in the common carotid artery (CCA) and ICA, as well as decreased resistance and increased flow velocity in the superficial temporal artery (STA) resulting from the effects of compensation (Ruan et al. 2006). The stenotic intracranial arteries commonly exhibit low or high flow velocity (Lee et al. 2004; Ruan et al. 2006). After direct bypass surgery, the patency of the STA graft is associated with increased flow velocity and decreased resistance in the STA on ultrasound (Wu et al. 2011); however, the exact changes in the ultrasound images after indirect revascularization surgery remain unclear.

Encephaloduroarteriosynangiosis (EDAS) surgery is one kind of indirect revascularization surgery, particularly for pediatric moyamoya patients using STA as a feeding artery. We hypothesized that the STA should exhibit significant changes on ultrasonography after EDAS compared with the pre-operative state. Thus, we compared ultrasonographic parameters between the pre- and post-operative states on the operated side to characterize serial changes in the extracranial and intracranial arteries after EDAS in moyamoya patients. In addition, because progression of MMD may confound the post-operative changes, we compared the hemodynamic parameters on the operated side with those for the contralateral non-operated side at serial time points.

METHODS

Patients

Between July 2012 and July 2014, consecutive pediatric moyamoya patients (<20 y of age) who had been diagnosed by cerebral angiography and were scheduled to undergo EDAS at the National Taiwan University Hospital were prospectively enrolled for this study. All but one patient had not undergone surgery for MMD before being enrolled; the exception had undergone EDAS on one side, and so this hemisphere was excluded from analysis because of a lack of pre-operative ultrasonographic data. All patients underwent follow-up angiography (cerebral angiography or magnetic resonance angiography) after EDAS to check for collateral establishment and disease progression. The demographic characteristics, neurologic events, angiographic findings and surgical complications of each patient were recorded. This study was approved by the Institutional Ethics Committee, and all enrolled patients and their parents gave informed consent.

Ultrasonographic assessment

In the present study, ultrasonographic examination was arranged before surgery and at 1, 3 and 6 mo afterward. The extracranial and transcranial ultrasound studies were performed using a color-coded ultrasound system (IE33, Philips Medical Systems, Bothell, WA, USA). These ultrasound examinations were performed by one neurologic ultrasound technician.

Extracranial ultrasound study. Extracranial arteries were assessed using an 11-3 MHz linear-array transducer. For the extracranial arteries, we measured peak systolic velocity, end-diastolic velocity, resistance index and flow volume of the CCA, ICA, external carotid artery (ECA) and STA. The ICA was measured 1.5–2 cm above the carotid bifurcation, and the ECA was assessed at the proximal ECA segment before the branching of the superior thyroid artery. The STA was analyzed at the common STA segment at the level of the ear. The flow volume of each vessel was calculated from the time-averaged mean flow velocity of the cross-sectional area of the individual vessel; this formula has been built into the ultrasound software. The mean flow velocity was calculated from the mean of all instant velocities within the spectrum over the heart cycle. In terms of the diameter of each vessel for calculation of cross-sectional area, we measured the diameter between inner luminal walls at end-diastole; we measured once for each vessel because pediatric patients were unable to tolerate prolonged examinations.

Transcranial color-coded sonography. Transcranial color-coded sonography (TCCS) was performed using a 5-1 MHz phased-array probe with angle correction. For the intracranial arteries, we measured peak systolic velocity, mean flow velocity and pulsatility index of the anterior cerebral artery (ACA), middle cerebral artery (MCA) M1 and M2 segments and posterior cerebral artery (PCA) on color Doppler images. The formula for determining mean flow velocity is built into the ultrasound software, and these data were automatically displayed. The highest flow velocity along the course of the individual vessel was recorded.

Statistical analysis

Categorical variables are presented as percentages, and continuous or discrete variables as means ± standard deviations. Student's paired *t*-test was used for univariate analysis between groups. First, we compared the hemodynamic parameters between the pre- and post-operative states. Cerebral hemispheres without complete post-operative follow up at these time points were excluded. Second, we compared the hemodynamic parameters on the operated side with those for the contralateral non-operated side after excluding patients who had undergone surgery on the contralateral side. Two-tailed p values < 0.05 were considered to indicate statistical significance. Data management and analysis were performed using Small Stata software (StataCorp LP, College Station, TX, USA).

RESULTS

Basic characteristics of these patients

Among the 15 patients enrolled in the study (Table 1), the mean age was 10.4 ± 4.6 y (range: 5–19 y), which included 11 males and 4 females. EDAS was performed on 19 cerebral hemispheres. Six of these also underwent indirect revascularization surgeries in addition to EDAS, including multiple-burr-hole surgery (MBH) (n = 3), encephaloperiosteosynangiosis (EPS) (n = 2) and encephalomyosynangiosis (EMS) (n = 1). The most common