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BRIEF COMMUNICATION

Immediate improvement of motor function after epilepsy surgery in congenital hemiparesis

***Tharick Pascoal, *Eliseu Paglioli, *André Palmini, *Rafael Menezes, and †‡Martin Staudt**

***Porto Alegre Epilepsy Surgery Program, Hospital São Lucas, Pontifical Catholic University of Rio Grande do Sul, Porto Alegre, Brazil; †Department of Pediatric Neurology and Developmental Medicine, University Children's Hospital, Tübingen, Germany; and ‡Clinic for Neuropediatrics and Neurorehabilitation, Epilepsy Center for Children and Adolescents, Schön Klinik Vogtareuth, Germany**

Summary

Hemispherectomy often leads to a loss of contralateral hand function. In some children with congenital hemiparesis, however, paretic hand function remains unchanged. An immediate improvement of hand function has never been reported. A 17-year-old boy with congenital hemiparesis and therapy-refractory seizures due to a large infarction in the territory of the middle cerebral artery underwent epilepsy surgery. Intraoperatively, electrical cortical stimulation of the affected hemisphere demonstrated preserved motor projections from the sensorimotor cortex to the (contralateral) paretic hand. A frontoparietal resection was performed, which included a complete disconnection of all motor projections originating in the sensorimotor cortex of the affected hemisphere. Surprisingly, the paretic hand showed a significant functional improvement immediately after the operation. This observation demonstrates that, in congenital hemiparesis, crossed motor projections from the affected hemisphere are not always beneficial, but can be dysfunctional, interfering with ipsilateral motor control over the paretic hand by the contralesional hemisphere.

KEY WORDS: Congenital hemiparesis, Sensorimotor reorganization, Epilepsy surgery, fMRI, Corticospinal tract.

Hemispherectomy often leads to a deterioration of contralateral hand functions. In some patients with congenital hemiparesis, however, motor functions of the paretic hand (contralateral to the operation) can remain unchanged after the operation, indicating that these functions were shifted to the contralesional hemisphere by the early unilateral brain lesion (van der Kolk et al., 2013). An immediate improvement of the paretic hand after a resection has, to date, never been reported.

Case Report

This 17-year-old boy with congenital left-sided hemiparesis had medically refractory seizures since the age of 3 years. The seizures started with a sudden inability to speak and a "sensation that the muscles of the left arm were tightened," followed by version of the head and eyes to the left, clonic movements of the left arm, and sometimes bilateral convulsions. Neurologic examination showed an alert, cooperative boy with left hemiparesis and hemianopia. His left limbs were spastic, although he could perform some finger movements (Video S1). Electroencephalography (EEG) showed bilateral sharp waves predominating in the right fronto-centroparietal regions, associated with irregular delta waves. Ictal patterns consisted of bilateral rhythmic discharges preceded by right frontocentral delta waves and, occasionally, low-amplitude polyspikes in both rolandic regions. Structural magnetic resonance imaging (MRI) showed a large cortico-subcortical lesion in the right hemisphere (Fig. 1A) and a small wedge-shaped ischemic lesion in the contralateral (left) parietal region, which did not manifest clinically (Fig. 1B). Functional MRI (1.5 T Siemens scanner, echo planar imaging (EPI), repetition time (TR) 0.6 msec, echo time (TE) 60 msec, scan time 3 s, slice thickness 4 mm) was acquired in a block design, with each functional study consisting of 60 scans alternating six periods of rest and six periods of activation. Data were realigned, smoothed (full-width half-maximum [FWHM] = 8 mm) and analyzed using Statistical Parametric Mapping (SPM2), with an activation threshold of $p < 0.0001$ (uncorrected). Repetitive grasping with the right (nonparetic) hand elicited activation in the central (rolandic) region of the left (contralesional) hemisphere (Fig. 1C); grasping with the left (paretic) hand elicited activation of the central area in the right (affected) hemisphere, without any additional suprathreshold activation (Fig. 1E). Epilepsy

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Address correspondence to Martin Staudt, Clinic for Neuropediatrics and Neurorehabilitation, Epilepsy Center for Children and Adolescents, Schön Klinik, Krankenhausstr. 20, D – 83569 Vogtareuth, Germany. E-mail: mstaudt@schoen-kliniken.de

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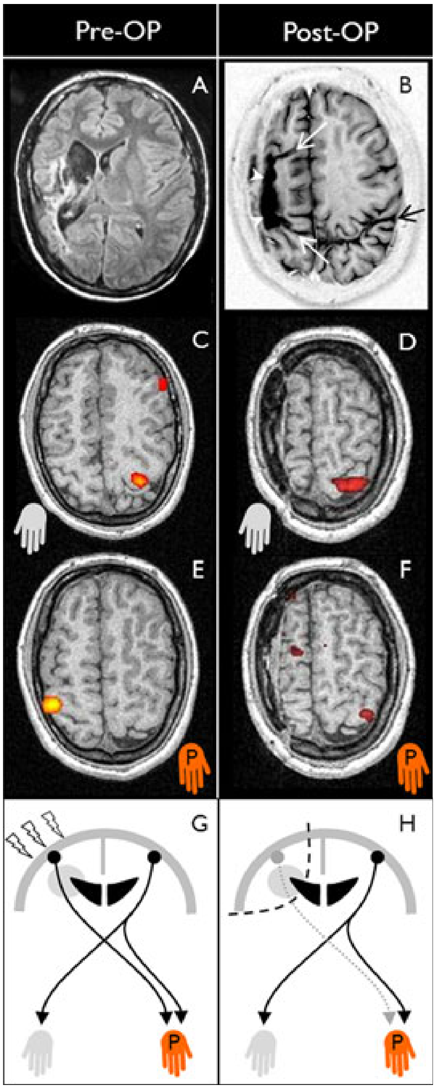

Figure 1.
Preoperative and postoperative structural/functional MRI and schematic illustration of corticospinal projections. (**A**) Preoperative axial fluid-attenuated inversion recovery (FLAIR) MRI depicting the right-sided cortico-subcortical infarction. (**B**) Postoperative inversion-recovery MRI depicting the surgery (corticectomy [white arrowheads]; frontal and parietal leukotomies [white arrows]) as well as the small additional parietal lesion in the "contralesional" left hemisphere (black arrow). (**C**–**F**) Functional MRI during unimanual grasping movements [**C**, **D**: nonparetic hand (symbolized in gray); **E**, **F**: paretic hand (symbolized in orange); **C**, **E**: preoperative fMRI; **D**, **F**: postoperative fMRI]. (**G**) Preoperatively, the paretic hand (P) received projections from both hemispheres. The epileptic dysfunction is symbolized by lightning bolts over the affected hemisphere (gray circle = lesion). (**H**) Postoperatively (black dashed line = resection), the crossed projection from the affected hemisphere has been disconnected (gray dotted line), so that the paretic hand receives input only from the contralesional hemisphere.
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surgery was performed at the age of 17 years. No changes in medication were made in the time around surgery; at that time, the patient had several seizures per day, with secondary generalization once per week. Electrical cortical stimulation of the rolandic cortex in the exposed right hemisphere consistently elicited motor activity in the left (paretic) hand and forearm with 4–6 mA and of the fingers of the paretic hand with 14 mA (Video S1). No motor activity was elicited with stimuli up to 14 mA in the other exposed cortical regions. Corticectomy was then performed, from the sylvian region to the midline, extending 8 cm from the frontal region until 5 mm anterior to Trolard's vein in the parietal region, thus including all aspects of the precentral and postcentral gyrus except for their most medial aspects. Following resection of this frontocentral "bloc," the lateral ventricle was opened, disconnecting the corona radiata and then sectioning the body and the isthmus of the corpus callosum. This left the frontomesial cortex in place, yet completely disconnected. Therefore, the entire rolandic cortex (precentral and postcentral gyrus) was either removed or disconnected. Postoperatively, an immediate and significant improvement of motor functions in the paretic hand was observed: Already on the first postoperative day, the patient was able to extend the fingers of the paretic hand much more easily, and spasticity had markedly decreased (Video S1). Postoperative functional MRI (fMRI) (13 months postoperatively) during active movements of the paretic hand elicited activation in the "hand knob" area of the contralesional hemisphere (Fig. 1F). At follow-up 3 years after the operation, the patient is still seizure-free, and manual abilities remain improved. Mirror movements were now tested, and were present both during voluntary movements with the paretic hand and with the nonparetic hand (Video S1). Cognitive functions had been normal preoperatively, and no change was observed after the operation.

Discussion

The most important observation in this patient was the immediate improvement of paretic hand function after the surgical removal and disconnection of the contralateral sensorimotor cortex. Taking the preoperative, intraoperative, and postoperative data together enables us to clarify the mechanism underlying this phenomenon.

Preserved active grasping after hemispherectomy in patients with congenital hemiparesis apparently depends on the presence of ipsilateral corticospinal projections from the contralesional hemisphere to the paretic hand (Rutten et al., 2002; Sun et al., 2009; Zsoter et al., 2012). Therefore, given the good postoperative function of the paretic hand in our patient, we can assume that he also possesses such ipsilateral corticospinal projections (Fig. 1G,H). Unfortunately, transcranial magnetic stimulation, the "gold standard" technique for the detection of such projections, was not available. The postoperative fMRI with activation in the hand area of the ipsilateral (contralesional) hemisphere, however, further corroborates this assumption (Fig. 1F).

In addition to these ipsilateral projections from the contralesional hemisphere, this patient also possessed contralateral corticospinal projections from the affected hemisphere to the paretic hand, as demonstrated by intraoperative electrical stimulation (Video S1). This bilateral type of corticospinal projections is not infrequent in patients with congenital hemiparesis (Staudt et al., 2002; Holmström et al., 2010). This constellation implies that, in the preoperative situation, the paretic hand received input from both the (affected) right hemisphere and the (contralesional) left hemisphere (Fig. 1G).

Postoperatively, when all motor projections from the affected hemisphere to the paretic hand had been disconnected, the paretic hand can receive only input from the (ipsilateral) contralesional hemisphere—and showed an immediate improvement (Video S1, Fig. 1H). This demonstrates that the projection from the (epileptic) contralateral hemisphere not only had not contributed to useful voluntary function, but had in fact worsened function, interfering with or inhibiting the ipsilateral control by the contralesional hemisphere. The dysfunctionality of the contralateral projection probably finds its explanation in the ongoing epileptic activity in the sensorimotor cortex of the affected hemisphere, which, after the operation, could no longer interfere with motor function of the paretic hand.

The interpretation of the preoperative fMRI is not as straightforward. The activation of the hand sensorimotor area in the affected hemisphere seems to suggest that active movements of the paretic hand at that time were predominantly controlled by the lesioned hemisphere, and not so much by the contralesional hemisphere. Although this might well be the correct interpretation, it could also be that this activation reflects only a preserved primary somatosensory (S1) representation of the paretic hand in the affected hemisphere (Thickbroom et al., 2001; Staudt et al., 2006). In fact, in this situation, the (contralateral) S1 activation is often stronger than the (ipsilateral) primary motor (M1) activation, which sometimes does not even reach the activation threshold (patient 4 in Staudt et al., 2006)—this would parallel the preoperative fMRI finding in the patient reported herein. Therefore, the question of whether the lesioned hemisphere was involved in voluntary movements of the paretic hand before the operation cannot be answered with the available data. Nevertheless, the different fMRI activation patterns nicely illustrate how the operation has changed motor control over the paretic hand.

In conclusion, this case report demonstrates that motor projections from an epileptic hemisphere can, at least in some patients with congenital hemiparesis, be harmful for the function of the paretic hand, and that surgical procedures that include the disconnection of these projections can "liberate" the paretic hand from this negative influence, leading to an immediate improvement of motor function. The detection of such projections should therefore not automatically be regarded as problematic in the planning of surgical procedures including their removal or disconnection, and patients with both contralateral and ipsilateral projections to the paretic hand could still be excellent candidates for hemispherectomies or similar procedures.

Disclosure

None of the authors has any conflict of interest to disclose. All authors confirm that they have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

References

Holmström L, Vollmer B, Tedroff K, Islam M, Persson JK, Kits A, Forssberg H, Eliasson AC. (2010) Hand function in relation to brain lesions and corticomotor-projection pattern in children with unilateral cerebral palsy. *Dev Med Child Neurol* 52:145–152.

Rutten GJ, Ramsey NF, van Rijen PC, Franssen H, van Veelen CW. (2002) Interhemispheric reorganization of motor hand function to the primary motor cortex predicted with functional magnetic resonance imaging and transcranial magnetic stimulation. *J Child Neurol* 17:292–297.

Staudt M, Grodd W, Gerloff C, Erb M, Stitz J, Krägeloh-Mann I. (2002) Two types of ipsilateral reorganization in congenital hemiparesis: a TMS and fMRI study. *Brain* 125:2222–2237.

Staudt M, Braun C, Gerloff C, Erb M, Grodd W, Krägeloh-Mann I. (2006) Developing somatosensory projections bypass periventricular brain lesions. *Neurology* 67:522–525.

Sun W, Fu W, Wang D, Wang Y. (2009) Ipsilateral responses of motor evoked potential correlated with the motor functional outcomes after cortical resection. *Int J Psychophysiol* 73:377–382.

Thickbroom GW, Byrnes ML, Archer SA, Nagarajan L, Mastaglia FL. (2001) Differences in sensory and motor cortical organization following brain injury early in life. *Ann Neurol* 49:320–327.

van der Kolk NM, Boshuisen K, van Empelen R, Koudijs SM, Staudt M, van Rijen PC, van Nieuwenhuizen O, Braun KP. (2013) Etiology-specific differences in motor function after hemispherectomy. *Epilepsy Res* 103:221–230.

Zsoter A, Pieper T, Kudernatsch M, Wilke M, Staudt M. (2012) Predicting hand function after hemispherotomy: TMS versus fMRI in hemispheric polymicrogyria. *Epilepsia* 53:e98–e101.

Supporting Information

Additional Supporting Information may be found in the online version of this article:

Video S1. This video shows the preoperative and postoperative hand function as well as the site and effect of intraoperative electrical stimulation. Note that the preoperative video depicts the typical level of hand function during that time, and not a temporary deterioration for example after a prolonged seizure.