Clinical Neurology and Neurosurgery 134 (2015) 126–129

Contents lists available at ScienceDirect

Clinical Neurology and Neurosurgery

journal homepage: www.elsevier.com/locate/clineuro

Case Report

# A possible variant of negative motor seizure arising from the supplementary negative motor area

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ARTICLE INFO

*Article history:*
Received 18 September 2014
Received in revised form 20 April 2015
Accepted 26 April 2015
Available online 6 May 2015

*Keywords:*
Negative motor area
Motor evoked potential
Ictal onset zone

## 1. Introduction

High-frequency electrical cortical stimulation of a certain cortical area in the frontal lobe produced "negative effects", arrest of voluntary movements without lack of consciousness; this cortical area has been termed "negative motor area (NMA)" [1,2].

Negative motor seizure (NMS) is a rare seizure that involves only the inability to conduct voluntary movements or praxis, without lack of consciousness [3]. To our knowledge, in only one report [3], by analyzing the features common to three cases, NMS were found to originate within the broad lateral and medial areas defined as NMA, but the specific electrodes showing the seizure-onset zone did not necessarily produce negative motor responses. NMA seems to be responsible for NMS, but the precise origin and mechanism of NMS has not been fully clarified.

Here we report a patient with a tumor in the medial right frontal lobe who manifested NMS as the sole seizure semiology, and who became seizure-free after resection of the tumor and adjacent NMA.

## 2. Case report

The patient was a 34-year-old, right-handed man. One year prior to referral to Kyoto University Hospital, he began experiencing brief stereotyped episodes in which he felt mirth without laughter for a few seconds while reading comic books whose contents were not funny. Six months later, he began having difficulty or slowing for a few seconds at a time while using both arms (e.g., while cleaning his body with a towel) or speaking in front of an audience, though others did not notice. None of these events devolved into positive motor seizures. Because the stereotyped episodes occurred repeatedly (once or twice a month), he consulted a neurosurgeon at his local hospital. His brain MRI revealed a tumor in the medial right frontal area, and the patient was referred to our hospital. Neurological examination was unremarkable, including lack of difficulty with alternating hand movements, which is typically seen in supplementary motor area (SMA) syndrome. MRI showed a T1-hypo-, T2-hyper-intensity lesion without gadolinium enhancement in the medial right frontal lobe anterior to the vertical anterior commissure (VAC) line (Fig. 1A). NMS in association with a low-grade tumor was suspected and 3-days

*Abbreviations:* NMS, negative motor seizure; NMA, negative motor area; SMA, supplementary motor area; VAC, vertical anterior commissure; EEG, electroencephalogram; ECoG, electrocorticogram; MEP, motor evoked potentials.

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http://dx.doi.org/10.1016/j.clineuro.2015.04.024
0303-8467/

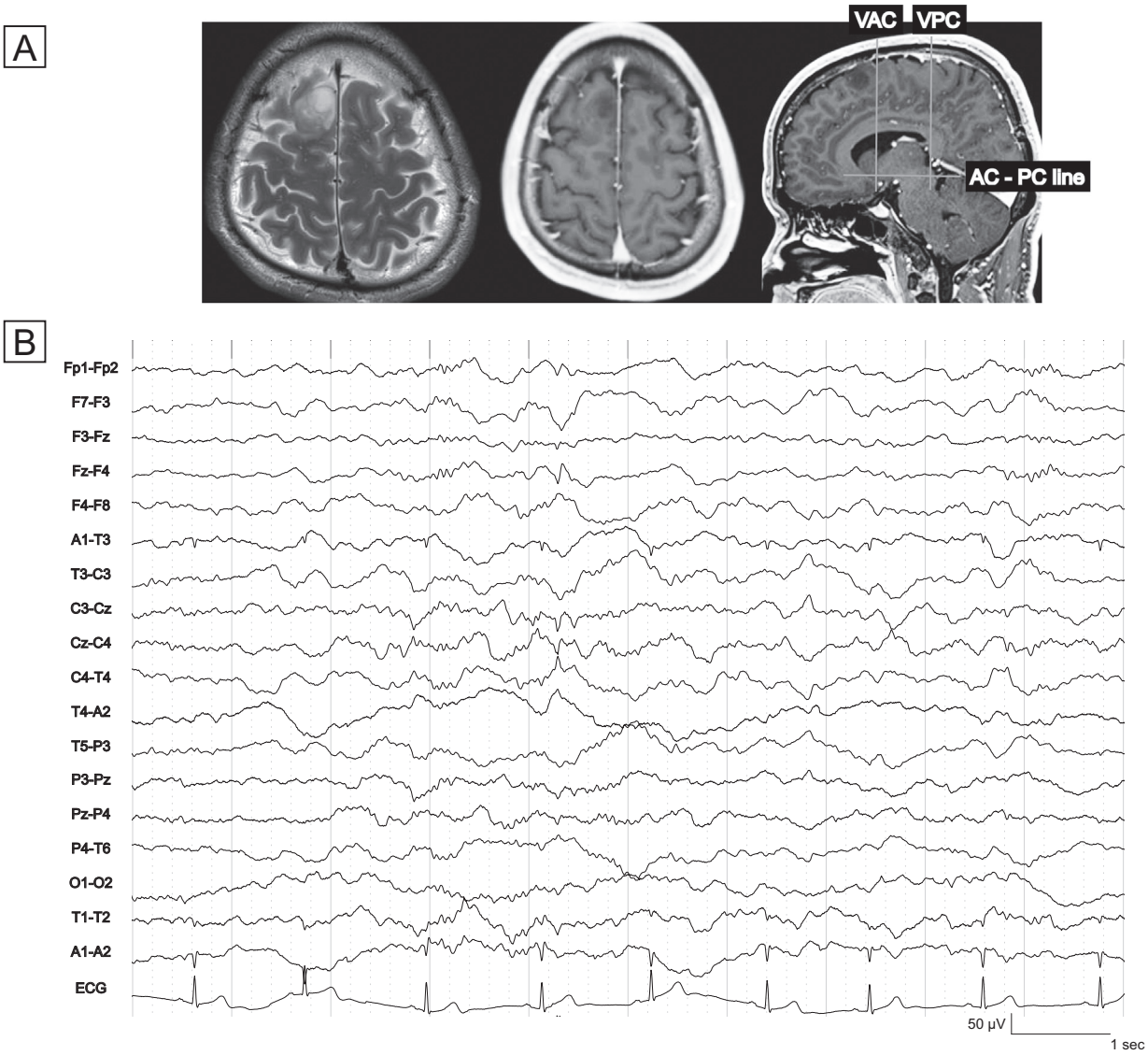


**Fig. 1.** Preoperative MRI data (A) and interictal electroencephalogram with scalp electrodes (B). A preoperative MRI shows a T1-hypo-, T2-hyper-intensity (A: left) tumor without gadolinium enhancement (A: middle and right) in the medial right frontal lobe anterior to the vertical anterior commissure line. Only a sharp transient is shown at the right fronto-central area with a maximum at C4 (B). AC–PC line: anterior commissure–posterior commissure line, VAC: vertical anterior commissure, VPC: vertical posterior commissure, ECG: electrocardiogram.

video-electroencephalogram (EEG) monitoring with scalp electrodes was performed. No habitual seizures were detected, and only a few sharp transients were recorded interictally at the right fronto-central area with a maximum at C4 (Fig. 1B).

The patient underwent awake craniotomy to probe motor function and identify a possible epileptic focus at or around the tumor. Two $4 \times 5$ subdural grid electrodes (recording diameter of 3 mm, inter-electrode distance 1 cm, Unique Medical, Tokyo, Japan) were placed over both the lateral and medial sides of the right frontal lobe. A 32-channel intraoperative monitoring system (MEE 1232 Neuromaster & MS 120B, Nihon-Koden, Tokyo, Japan) was used both for recording raw electrocorticogram (ECoG) data and delivering electrical stimuli. ECoG data was digitized with a sampling rate of 5000 Hz and a band-pass filter of 0.5–1500 Hz.

To avoid the risk of evoking seizures, functional motor mapping was performed in the awake state by applying single-pulse electrical cortical stimulation (pulse width of 0.3 ms, 1 Hz, alternating polarity, 10–15 mA) and recording motor evoked potentials (MEPs), as reported elsewhere [4]. Surface electromyograms were recorded from the bilateral deltoid, extensor carpi radialis, abductor pollicis brevis and tibialis anterior muscles. MEPs were recorded in the resting condition and during adequate muscle contractions at the same stimulus intensity. In electrodes where MEPs were not elicited by 1-Hz electrical cortical stimulation, 5-train electrical cortical stimulation (a square pulse of 0.2 ms with 2 ms interval, 10–15 mA) was applied to identify motor function [5]. Functional motor mapping was performed only for the clinical purpose, and the electrical cortical stimulation was approved by the Kyoto University Graduate School and Faculty of Medicine, Ethics Committee (IRB C573).

Since we had not performed intraoperative MRI, the placement of electrodes was identified based on operative observation and neuro-navigation data.

In the awake state, frequent interictal spikes were focally recorded posterior to the tumor in the medial frontal area (Electrodes B09, 13, 14, and 19 in Figs. 2 and 3A). Spikes occurred most frequently at Electrode B09. No spikes were shown on the lateral frontal cortex. Based on MEP patterns elicited by 1-Hz electrical cortical stimulation as defined previously [4] (e.g., the response of the bilateral upper extremities or contralateral lower extremity), the SMA was defined at the electrode pairs of B01-02, B06-07, and