

**Awake surgery for a violin player: monitoring motor and music performance, a case
report**

Running title: monitoring motor and music performance in awake surgery

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Abstract

Objective: We report the case of a professional violin player who underwent an awake craniotomy to resect a tumour in the left supplementary motor area, an area involved in motor planning. **Method:** A careful pre- and intraoperative monitoring plan for music performance and complex motor function was established that could be used in combination with cortical stimulation. **Results:** The patient suffered an epileptic seizure during cortical stimulation. The monitoring of complex motor and musical functions was implemented with the patient playing the violin while the resection was performed. Almost complete resection was achieved with no notable postoperative deficits contributing to functional impairment .

Conclusions: The multidisciplinary approach, involving neurosurgery, neuropsychology, anaesthesiology, and clinical neurophysiology, allowed us to successfully cope with the theoretical and practical challenges associated with tailored care for a professional musician. The music and motor monitoring plan is reported in detail to enable other sites to reproduce and adapt it accordingly.

Keywords: craniotomy, direct electrical stimulation, functional mapping, musical ability

Introduction

The supplementary motor area (SMA) is a brain area involved in the control of movement. Early evidence on the function of the SMA was provided by Penfield and Welch (1951), who described the types of responses following direct cortical electrical stimulation (DCS) to this area. Vocalizations with complex patterns (in rhythm, volume, pitch) were elicited, also accompanied by face and jaw movements. Speech, for example during counting or word repetition, could be inhibited for the duration of the stimulation while the patient remained conscious. Regarding the extremities, DCS elicited arm movements as part of complex motor responses, also involving for example leg, eye, or head movements. These movements of the extremities followed three main patterns: a slow movement reaching an end-point akin to a posture, a series of movements following a complex pattern, and rapid uncoordinated movements. Resection of the SMA may produce the so-called SMA syndrome (Laplaine, Talairach, Meininger, Bancaud, & Orgogozo, 1977). The syndrome is characterised by a transient akinesia of the limbs contralateral to the resected hemisphere with (nearly) complete recovery of spontaneous movements within a few weeks or months.

Playing a music instrument requires the coordination of bimanual movements and motor sequencing with the right timing, among other processes. Evidence suggests an important role for a number of structures in music performance (see for reviews Goldberg, 1985; Zatorre, Chen, & Penhune, 2007). Neuroimaging and neuropsychological studies have linked the ability to time movement with precision to the cerebellum, basal ganglia, SMA, and premotor cortex. In addition, the ability to sequence movements has also been linked to these structures. Thus, the SMA and the premotor cortex are important cortical structures involved in the complex motor abilities required for music performance.

Case report

A professional violin player, aged between 35-40 with more than 16 years of education

consulted the neurologist after two seizures. The patient was left-handed, according to the Edinburgh Handedness Inventory (Oldfield, 1971). No neurological deficits were found. Magnetic resonance imaging revealed a lesion in the left SMA region, suspected to be a low grade glioma (Fig. 1A). Given the location of the patient's tumour and the importance of this brain region to various aspects of motor behaviour that are necessary for music performance, careful pre- and intraoperative monitoring plans were established.

As part of the preoperative procedure, functional magnetic-resonance (fMRI) and tractography data were acquired. The fMRI protocol consisted of a left- and right-finger tapping task ("motor"), a word-reading task ("speech"), and a verb-generation task ("language"). The motor tasks evoked expected activations of the corresponding (right vs left) pre- and postcentral gyri, but no activations were observed either in the SMA or infiltrating the tumour. For speech, activation was observed in the left supramarginal gyrus and asymmetrical (left > right) activation in the pre- and postcentral gyri. Weak activation in the left SMA was also observed, but that did not reach the tumour. For language, activation was observed in the left inferior and middle frontal gyri, and left supramarginal gyrus, and asymmetrically (left > right) in the prefrontal cortex. Activation in the right middle temporal gyrus was also found. Finally, bilateral SMA activation was observed infiltrating the tumour. This pattern of activation suggests a relatively left-hemisphere dominance for language function, although some right-hemisphere involvement is likely (Szaflarski et al., 2002). Tractography indicated that the tumour was near the corticospinal tract and the arcuate fasciculus (Fig. 1B).

Given the activation patterns for speech and language in the vicinity of the tumour, as well as the potential criticality of the SMA for playing the violin, it was decided to use awake monitoring of motor function, speech, language, and music during the neurosurgical procedure.

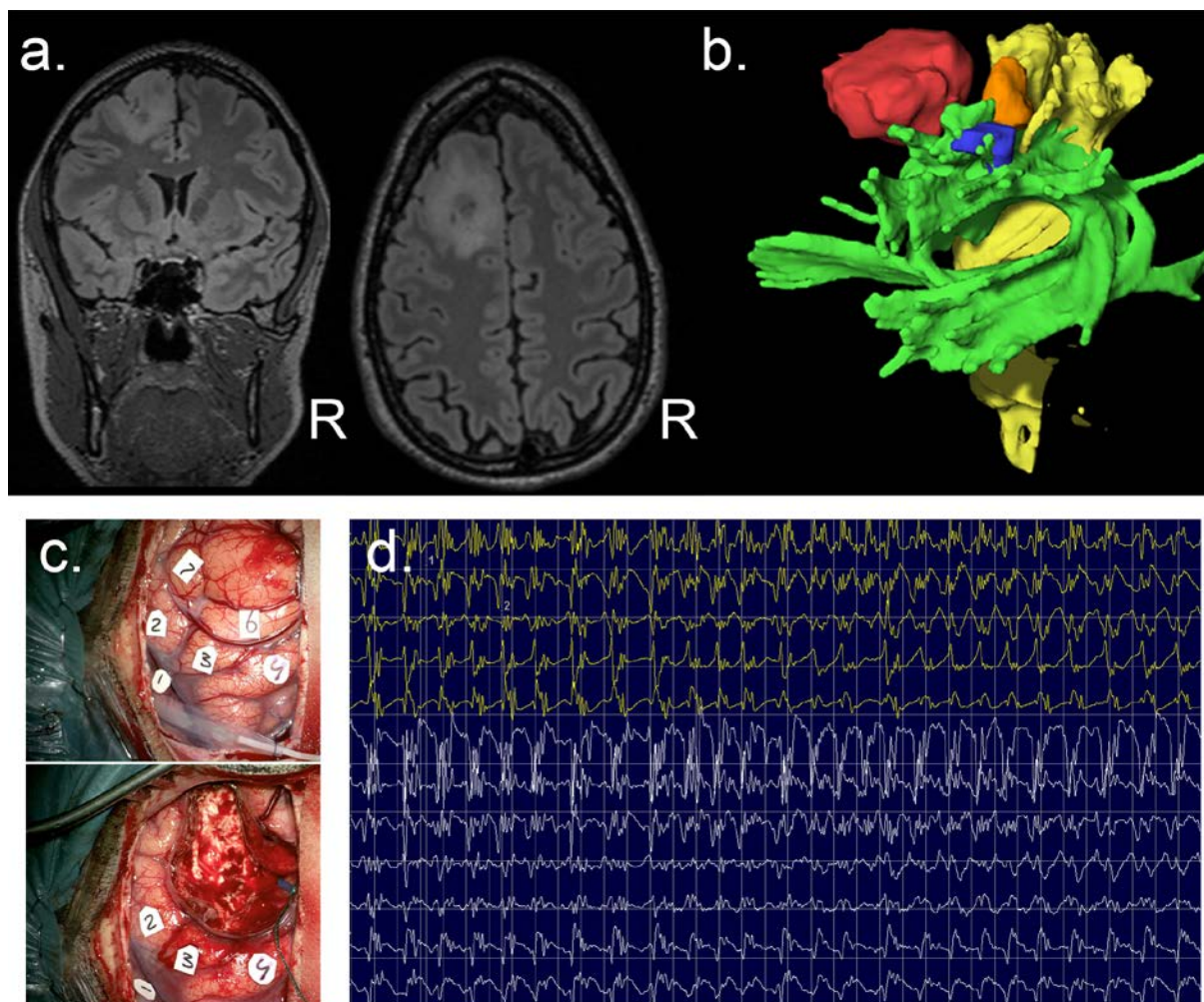


Figure 1. **a.** Flair images of the left-hemisphere lesion in the supplementary motor area. R = right. **b.** Lateral view of 3D reconstructed model for surgical planning with the tumour in red, hand function in orange, lip function in blue, corticospinal tract in yellow, and arcuate fasciculus in green. **c.** Intraoperative photograph of the cortical surface before (upper) and after (lower) the resection. 1 = precentral gyrus, hand area; 2, 3, 4 = precentral gyrus, from the lateral (2) into the medial (4) direction; 6, 7 = tumour, from the lateral (7) into the medial (6) direction. **d.** Elocorticography recording of epileptiform discharges via an 8-contact electrode strip following 4 mA stimulation with the Penfield technique. The upper five tracings (yellow) are in bipolar montage (contacts: 3-4, 4-5, 5-6, 6-7, and 7-8) and the lower seven tracings (white) are in referential montage (contacts: 1, 3, 4, 5, 6, 7, and 8). Electrode 2 did not make good contact with cortex and is therefore not shown. Generalised discharges in the form of repetitive spike-wave and polyspike-wave complexes at approximately 3 Hz are seen on all contacts.

Preoperative cognitive, language, motor, and musical abilities

Apart from the seizures and the normal very recent distress reaction to the news of the tumour, patient nor partner reported cognitive or behavioural changes. Extensive preoperative neuropsychological testing was conducted to objectively measure the patient's cognitive

abilities. Neuropsychological tests were chosen to cover different cognitive domains, including abstract verbal and visual reasoning, working memory and episodic memory, executive functioning, attention, language (comprehension, naming, and fluency), and visuoconstruction (Bouma et al., 2012, Lezak, 2004). As can be seen from Table 1, none of the test results indicated any impairments (i.e., as classified by 2 standard deviations below the age- and education-adjusted normative mean as cut-off score, T score < 30, percentile < 2.3). In addition, the Dutch version of the Hospital Anxiety and Depression Scale (HADS) was administered to assess the experience of anxiety and depressive complaints (Spinhoven et al., 1997). The low scores indicated low levels of experienced anxiety and depressive states. Psychological wellbeing was assessed with the 90-item revised Dutch version of the Symptom Checklist (Arrindell et al., 2003). The SCL-90-R is a multidimensional self-report questionnaire designed to screen for a broad range of psychological problems containing the symptom dimensions agoraphobia, anxiety, depression, somatisation, cognitive performance difficulty, interpersonal sensitivity, anger-hostility, and sleep disturbance. The patient reported no elevated symptoms in any of those dimensions.

In addition, complex motor function and musical abilities were tested comprehensively. Until recently, standardised ways of assessing sensorimotor function pre-, intra-, and postoperatively had not been reported. Recently, a standardised protocol has been proposed to fill this gap (Becker et al., 2016). Regarding motor function, this protocol consists of asking the patient to execute a number of movements following verbal commands. The target movements range from facial (e.g., blink eyes, wrinkle nose), to hand and arm, leg and foot. For the hand and arm, commands include “shake hands” and “rotate wrist”, whereas fine hand motor function is probed by finger tapping and “rock-paper-scissors”. For the leg and foot, commands include “lift knee” and “move toes”. A standardised rating scale accompanies the protocol to classify the amount of impairment.

We based our assessment on this recently developed protocol and expanded it to further include the (rhythmic) change of motor configurations. Moreover, a protocol was developed to assess musical abilities. The available literature provides little detail on the protocols used to monitor motor function and musical abilities pre- and intraoperatively. Therefore, we describe the procedure we adopted in detail below as it could be useful in guiding the monitoring plan in other sites.

The tasks we employed were designed to span a range of simple to complex finger and hand movements and simple to complex musical abilities (including melody and rhythm). Rhythm, i.e., regular pace, in the execution of all movements was emphasised. The following motor tasks were included:

- 1) Hand configuration: based on “rock-paper-scissors” (Becker et al., 2016), the patient alternated between two hand configurations (e.g., rock-paper-rock-paper) at a regular pace for six seconds, first each hand individually and then simultaneously. Three different pairs of configurations were chosen (rock-paper, paper-scissors, rock-scissors).
- 2) Finger tapping: the patient performed index-finger tapping on the keyboard at a regular pace.

The remaining tasks all included a music component of varying difficulty. Two tasks were performed both on the keyboard and on the violin (3 and 4). For the violin, the use of both bow strokes and pizzicato was assessed:

- 3) Major scale
 - For the violin, two octaves of G-major
 - For the keyboard, G to D, one different finger per note, each hand individually and then both hands simultaneously

- 4) Major arpeggio
 - For the violin, two octaves of G-major
 - For the keyboard, two octaves of C-major, one different finger per note, each hand individually and then both hands simultaneously
- 5) On the keyboard, the patient played intervals (C, E, G, F, D, C) repeatedly, one different finger per note, each hand individually.
- 6) Solfège: five pieces of increasing rhythmic and melodic complexity were used
- 7) On the violin, the patient played two easy pieces by heart: “Happy birthday”, “Frère Jacques”
- 8) On the violin, the patient performed sight-reading (i.e., the patient had never seen the piece before) of a relatively difficult piece, Telemann's Suite in A minor, TWV 55:A2, *Overture*

The hand configuration task (Task 1 above) was performed first. Then, all tasks using the keyboard were performed (in the order mentioned above). The solfège followed and finally all tasks using the violin were performed. A trained musician (first author) assessed the patient's performance. No deficits were noted in any of the tasks.

In addition, the patient chose a relatively complex, but familiar piece of music for playing by heart during the surgery (Bach's Partita No. 2 in D minor, BWV 1004, *Allemande*) and was asked to get familiarised at home with playing lying down. The familiarity was important so that baseline performance was guaranteed to be flawless. The practice at home was encouraged so that the intraoperative performance would be as much as possible only dependent on the direct cortical stimulation and resection, rather than on the awkwardness of the position. As reported below, this turned out to be an important part of the monitoring plan as none of the above tasks could be performed intraoperatively.

Intraoperative procedure

A scalp block was placed using ropivacaine under dexmedetomidine and low-dose propofol sedation. Further sedation was exclusively performed with dexmedetomidine in dosages adapted to the requirements of the surgical intervention. The exposed cortical surface can be seen in Figure 2A before and after the resection.

The central sulcus was identified using the phase reversal of the somatosensory evoked potential recorded with an eight-contact electrode strip during right median nerve stimulation. After that, the strip was used for electrocorticography. Monopolar anodal train-of-five stimulation (inter-stimulus-interval = 2 ms, pulse width = 0.5 ms) was used to map motor function in the precentral gyrus. At a stimulation intensity of 4 mA a reproducible motor evoked potential was found in the m. flexor carpi radialis and m. abductor digiti minimi. Classical bipolar Penfield stimulation (50 Hz, 4 seconds duration) was used to perform speech and language mapping during a picture naming task. Stimulation intensity was gradually increased with steps of 1 mA starting from 1 mA. The patient developed after-discharges following 4 mA stimulation of site 3 (see Figure 2A), which rapidly (within seconds) spread into a generalised seizure (Figure 2B). Rapid application of cold Ringer's lactate solution to the cortex was not effective, so a bolus of propofol (50 mg) and clonazepam (1 mg) were given, which successfully terminated the seizure as confirmed by the electrocorticography. Consequently, language mapping by means of the picture naming task was stopped. We waited for 45 minutes after the seizure stopped. No further epileptiform discharges were seen, but the patient was not fully awake. Therefore any residual benzodiazepines were counteracted by giving a bolus of flumazenil. The patient regained full consciousness, which was confirmed by adequate picture naming for a small number of trials.

Because of the generalised seizure at a relatively low current (while no deficit was found yet), it was decided not to use direct cortical stimulation anymore, since it was expected

that a subsequent seizure would make the monitoring impossible and potentially cause airway obstruction. Therefore, the prepared protocol could not be used as planned. It was decided to continue with tumour resection guided by neuronavigation while having the patient playing by heart the piece of music that was chosen and practised while lying down at home. A trained musician (first author), who assisted in the awake procedure, monitored the patient's performance throughout the resection. Given that the preoperative performance of this piece was flawless, the intraoperative performance was monitored for any type of deviation from the expected. During the resection, a musical mistake was noted only once and approximately 95% of the tumour could be resected.

Postoperative outcome

Postoperative days were uneventful except for a slight transient dyscalculia. The patient was discharged at day 4. Postoperative MRI scan showed slight residual increased FLAIR signal at the ventromedial border of the resection. Histopathological diagnosis showed an oligodendroglioma, WHO grade II, molecular analyses confirmed complete loss of 1p and 19q arms and an IDH1 mutation. No adjuvant therapy was administered because of the low risk profile (age <40, KPS > 70, gross total resection and oligodendroglioma diagnosis).

After two weeks, the patient resumed most activities. Six weeks after the surgery, the patient resumed playing with the philharmonic orchestra. The patient also started a new and highly cognitive-demanding job, involving communicative, organisational, and planning skills. Three and six months after the surgery, the suspected residual was unchanged. Upon the patient's request, no neuropsychological assessments were performed postoperatively.

Discussion

A number of limitations of this report are worth mentioning. Foremost, the patient's postoperative profile was not based on neuropsychological assessments, but rather only on self-reported function. In this regard, we cannot be certain that cognitive impairments were

entirely absent. However, with respect to the musical abilities, it is likely that any impairments would have prevented the patient from playing professionally. The fact that the patient resumed the professional musical activities soon after the surgery is an indication that no deficits were present contributing to notable functional impairment. Another important issue comes from the fact that we could not test our complex-motor and music protocol intraoperatively during DCS. However, the tasks proved suitable for the preoperative assessment. Moreover, given that the protocol was designed with the intraoperative setting in mind, we believe it has the potential to guide monitoring during DCS. The level of detail reported here should enable other clinicians to reproduce and adapt the procedure accordingly.

In summary, we found no impairments in motor, cognitive, or musical abilities in our patient due to a tumour infiltrating the left SMA preoperatively. After left-SMA resection, no deficits were noted contributing to functional impairment. The procedure adopted relied on a close collaboration between the different disciplines, i.e., neuropsychology, music theory, neurosurgery, anaesthesiology, and clinical neurophysiology. This multidisciplinary approach allowed us to successfully cope with the theoretical and practical challenges associated with tailored care for a professional musician.

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Table 1. Preoperative neuropsychological assessment.

Test	Standard score	Classification
<i>Abstract reasoning</i>		
WAIS-IV, similarities	12 (ss)	Average
WAIS-IV, matrix	12 (ss)	Average
<i>Working Memory</i>		
WAIS-IV, digit span, tot	11 (ss)	Average
WMS-III, spatial span, tot	13 (ss)	Average
<i>Processing speed</i>		
WAIS-IV, coding	10 (ss)	Average
<i>Executive functioning/Attention</i>		
Trail Making Test, B/A	55 (T score)	Average
BADS ¹ , zoo map, cond1+2	3 (profile score)	no deficit
Brixton Spatial Anticipation Test ²	30-40 (perc)	Average
Stroop Color Word test; T score is inference score	41 (T score)	Average
<i>Episodic memory</i>		
RAVLT ³ , mean tot score trials 1-5	41 (T score)	Average
RAVLT, delayed recall score	42 (T score)	Average
LLT-R ⁴ , mean nr errors 1-5	36 (perc)	Average
LLT-R, nr errors delayed	50 (perc)	Average
WMS-IV, logical memory imm	16 (perc)	Average
WMS-IV, logical memory delayed	25 (perc)	Average
<i>Language</i>		
Letter fluency (tot trials D-A-T)	48 (T score)	Average
Category fluency (tot trials)	57 (T score)	Average
Boston naming test	10-25 (perc)	(below) average
Token Test, short		no deficit
<i>Visual Construction (+memory)</i>		
ROCF (copy)	Perc >16	Average
ROCF (immediate recall)	Perc. 62	Average
ROCF (delayed recall)	Perc. 69	Average

Note: perc = percentile; ss = standard score; tot = total; sec = seconds; nr = number; imm = immediate; cond = condition; WAIS = Wechsler Adult Intelligence Scale; WMS = Wechsler Memory scale; BADS = Behavioural Assessment of the Dysexecutive Syndrome; RAVLT = Rey Auditory Verbal Learning Test; LLT-R = Location Learning Test-Revised; ROCF = Rey Osterrieth Complex Figure test. Education level (6) was administered in agreement with the Dutch educational system (1=less than primary school; 7=academic degree, Verhage, 1964). Premorbid intellectual level was estimated with the Dutch version of the National Adult Reading Test and indicated an average intelligence matching patient's obtained educational level. ¹Wilson, B. A., Alderman, N., Burgess, P. W., Emslie, H., & Evans, J. J. (2003). Behavioural assessment of the dysexecutive syndrome (BADS). *Journal of Occupational Psychology. Employment and Disability*, 5, 33-37. ²Burgess, P. W., & Shallice, T. (1997). *The Hayling and Brixton Tests*. Burry St. Edmunds, UK: Thames Valley Test Company. ³Van Der Elst, W., Van Boxtel, M. P. J., Van Breukelen, G. J. P., & Jolles, J. (2005). Rey's verbal learning test: normative data for 1855 healthy participants aged 24–81 years and the influence of age, sex, education, and mode of presentation. *Journal of the International Neuropsychological Socie-*

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