

# Musical hallucinations following insular glioma resection

## Case report

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Hallucinations can be auditory, visual, tactile, gustatory, or olfactory, and can be caused by psychiatric (such as schizophrenia and depression), neurological (such as cerebrovascular accidents, neoplasia, and infection), or endocrine and metabolic disorders. Musical hallucinations related to neurological disorders are rare. The authors present a case of a patient with a right insular glioma who developed transient musical hallucinations after microsurgical resection of the tumor. (DOI: 10.3171/2009.12.FOCUSFOCUS09243)

**KEY WORDS** • insula • musical hallucination • glioma

**H**ALLUCINATIONS are perception disorders characterized by the sensation of an external stimulus without the presence of such a stimulus. The term hallucination was introduced in the psychiatric literature in 1837 by Esquirol.<sup>23</sup> Hallucinations can be auditory, visual, tactile, gustatory, or olfactory, and can be caused by psychiatric (such as schizophrenia and depression), neurological (such as cerebrovascular accidents, neoplasia, and infection), or endocrine and metabolic disturbances.<sup>1-4,6,11-15,17-20</sup>

Music and neuroscience have been related for a long time. In 1977, Critchley and Henson published the book *Music and the Brain*. Despite countless advances in this field during the last 30 years, *Music and the Brain* remains one of the main books on this topic.<sup>23</sup> Auditory hallucinations in which music is perceived are rare and occur mainly in cases of deafness or psychiatric disorders.<sup>1</sup> A small number of cases are related to neurological disorders, generally due to alterations of the primary auditory area.<sup>3,4,17,18</sup>

Insular gliomas are usually slow-growing tumors of low-grade malignancy.<sup>25-27</sup> They may grow large before a diagnosis is made.<sup>25-27</sup> The main symptoms of these tumors are complex partial seizures with or without secondary generalization.<sup>25-27</sup> Due to the deep localization and the close relationship with the MCA, tumor resection in insular topography is considered a highly complex surgical procedure.

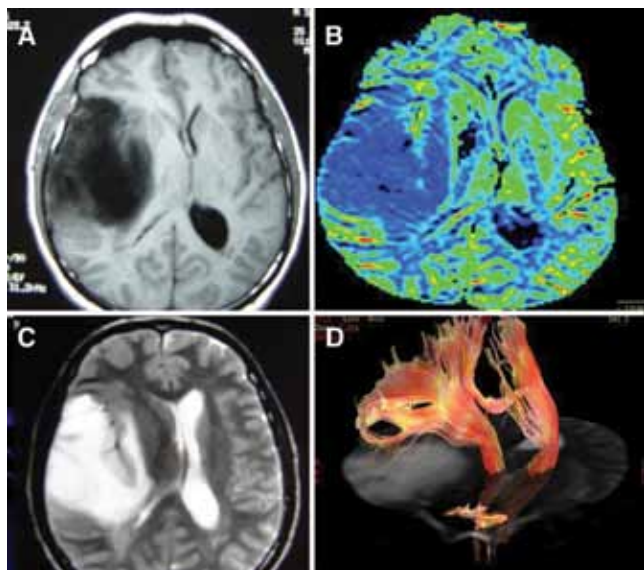
In this paper we report the case of a patient who underwent microsurgical resection of a large right insular tumor and who developed musical hallucinations of limited duration during the early postoperative period. As far as we know, this is the first report of a patient with musical hallucinations after insular tumor resection.

## Case Report

**History and Presentation.** This right-handed 34-year-old man was first seen at our institution due to new onset of episodic intense headaches and 1 episode of complex partial seizures. The neurological examination was unremarkable. A CT scan and 1.5-T MR imaging with Gd (T1-weighted, T2-weighted, FLAIR, perfusion, and tractography) disclosed a nonenhanced intraaxial tumor in the right insula, extending to the superior temporal gyrus and enveloping the planum polare and planum temporale (Fig. 1).

**Operation and Postoperative Course.** The patient underwent resection of the tumor (> 90%) using microsurgical techniques, and underwent monitoring for somatosensory evoked potentials and transcranial electric motor evoked potentials. Pathological examination of the tumor specimen revealed a WHO Grade II glioma (Fig. 2). The surgical approach was conducted via a right frontotemporal craniotomy, using an interfacial dissection of the temporal fascia and microdissection of the sylvian fissure (transsylvian approach).<sup>16</sup> The tumor was aspirated

Abbreviations used in this paper: MCA = middle cerebral artery.



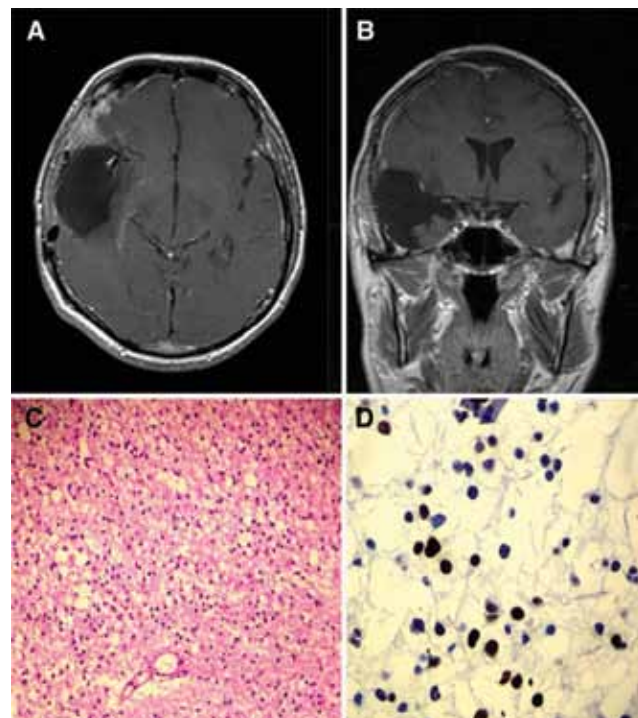
**Fig. 1.** Preoperative MR images of the patient. **A:** Contrast-enhanced T1-weighted image showing a nonenhanced lesion in the right insula causing midline deviation. **B:** Perfusion-weighted image showing the lesion. **C:** Contrast-enhanced T2-weighted image showing the absence of an important vasogenic edema. **D:** Diffusion imaging tractography showing the relation of the corticospinal tract to the tumor.

between the M<sub>2</sub> and M<sub>3</sub> segments of the MCA. The tumor portion extending to the planum polare and planum temporale was resected through the inferior circular sulcus of the insula, and a transopercular approach was used at the superior temporal gyrus, which was necessary to resect tumor in the lateral part of the Heschl gyrus. Deep resection of the tumor close to the internal capsule was stopped when there was an alteration in the intraoperative motor evoked potential.

The patient did not develop any neurological deficit as a result of the surgery. Diphenylhydantoin was initiated in the immediate preoperative period as prophylaxis for seizures. After 1 month, and while still receiving 100 mg of diphenylhydantoin three times a day, the patient began to have musical hallucinations. These hallucinations were characterized by a song that was very familiar to the patient although he was not able to identify it. The hallucinations occurred several times during the day, lasted several seconds, and were not associated with alterations of consciousness. This phenomenon occurred daily for approximately 2 months, and then disappeared. An electroencephalogram obtained at this time revealed discharges in the right centroparietal region and in the temporal regions of the right hemisphere. The patient has been asymptomatic for 1 year after surgery.

## Discussion

Musical hallucinations can be observed to occur in association with otologic, psychiatric, and neurological causes, or their combination. Sporadic cases have been reported in patients with psychiatric, otorhinolaryngologic, or neurological problems.<sup>1-4,6,11-15,17-20</sup> It has been assumed that the upper portion of the temporal lobe is



**Fig. 2.** Postoperative contrast-enhanced MR images (**A and B**) and photomicrographs (**C and D**). **A:** Axial T1-weighted image obtained 3 months after surgery showing tumor resection (> 90%) with a small remaining tumor in the posterosuperior region of the tumor cavity. **B:** Coronal T1-weighted image showing the tumor resection. **C:** Tumor specimen showing features of a WHO Grade II astrocytic neoplasm. H & E. Original magnification  $\times 100$ . **D:** Immunohistochemical specimen showing < 2% staining for Ki 67 (MIB-1). Original magnification  $\times 400$ .

the etiopathogenic substrate of this phenomenon. The functional imaging studies using the paradigm “listen to music” result in a bilateral activation of the brain.<sup>21,23</sup> On the other hand, studies of patients with acquired amusia suggest that the structures involved in this activity are the superior temporal cortex, the insula, and the frontal lobe of the nondominant cerebral hemisphere.<sup>23</sup> Although less common, lesions in the dominant cerebral hemisphere can also cause deficits in musical perception. For the latter aspect, however, we should consider the bias that patients with lesions in the left hemisphere generally present with aphasia, which makes it more difficult to perform nonlinguistic ability tests.<sup>23</sup>

Musical hallucinations can involve 3 large groups of alterations: otologic, psychiatric, and less commonly, neurological alterations. Regarding this last group, our case is the first report of musical hallucinations involving resection of a tumor in the insula topography.

Musical hallucinations associated with deafness have an estimated prevalence of approximately 2.5% in older patients.<sup>6</sup> Usually patients state that they hear familiar sounds, such as popular music.<sup>13,24</sup> Sometimes a humming sound precedes the musical hallucination. Hearing musical instruments and voices is a common phenomenon. Based on the humming that developed into musical hallucinations, Gordon<sup>12</sup> proposed that the etiological basis of the phenomenon is in the cochlea, although it is impos-

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sible to confirm that the mechanism has been explained based only on this premise. The release hallucinations concept is based on West's perceptive theory, whose main hypothesis is that blindness and deafness interrupt external sensorial stimuli necessary to inhibit brain memory evocation.<sup>5,7,22,23</sup> This phenomenon will release previously stored perceptions and visual or auditory memories.<sup>3,4</sup> Release musical hallucinations occur mainly in the elderly with hearing deficiencies, mainly while awakening. The atypical antipsychotic drug quetiapine improved these symptoms in some patients.<sup>7</sup> A similar phenomenon could be the reason for the transient symptoms experienced by our patient; the surgical procedure may have been responsible for the transitory release of the primary auditory area. Musical hallucinations associated with psychiatric disease are less common than verbal auditory hallucinations observed in schizophrenic, depressed, obsessive-compulsive, and alcoholic patients. Musical hallucinations are generally considered rare in psychiatric patients.<sup>3</sup> Fukunishi et al.<sup>11</sup> identified only 6 patients with musical hallucinations (3 of whom were deaf) in a population of 3578 psychiatric patients. However, this estimate may not be accurate, because Hermesh et al.<sup>14</sup> identified 20% of 190 psychiatric patients (mainly patients with obsessive-compulsive problems) who developed musical hallucinations at some period in their life.

Musical hallucinations associated with neurological diseases are less frequent than in the 2 former groups. In case reports, they are usually associated with abnormalities of the right nondominant cerebral hemisphere. Although quite rare, there are reports of musical hallucinations associated with dementia, meningioma,<sup>19</sup> gliomas,<sup>9</sup> subarachnoid hemorrhage,<sup>20</sup> cerebral ischemia,<sup>2</sup> and pontine hemorrhage.<sup>18</sup> To the best of our knowledge, there are no previous reports of hallucinations associated with insular gliomas.

On the other hand, the auditory aura is a rare phenomenon. In the series of 8000 patients of Florindo et al.<sup>10</sup> the auditory aura was present in only 121 cases. The role of laterality in epilepsy, where aura musical hallucinations occur, is a mechanism that must still be elucidated. The clinical characteristics of the case reported, in association with the electroencephalographic findings and the resection of the tumor tissue at the level of the planum temporale (more specifically, the anterior transverse temporal gyrus [Heschl gyrus]), lead us to believe the hypothesis that the postoperative musical hallucinations experienced by our patient could be a simple partial seizure due to surgical manipulation of the planum temporale or a release phenomenon.

Insular gliomas are tumors that become very large in many patients before they become symptomatic.<sup>25-27</sup> Sometimes these tumors are a casual finding observed during MR imaging. The cortex of the insula is an interface between the allocortex and the neocortex and constitutes part of the paralimbic system. It is related to motor, sensorial, language, auditory-vestibular, and cognitive functions.<sup>8</sup> The surgical treatment of these lesions is technically challenging because of their proximity to the internal capsule and the close anatomical relationship with the segments of the MCA and its branches.<sup>25-27</sup> The

MR imaging tractography performed in this case illustrates the close proximity of the deep tumor portion to the corticospinal tract.

### Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Conception and design: GR Isolan. Acquisition of data: GR Isolan, JA Bragatti, C Torres. Analysis and interpretation of data: GR Isolan, MM Bianchin. Drafting the article: GR Isolan, MM Bianchin. Critically revising the article: MM Bianchin, G Schwartzmann. Reviewed final version of the manuscript and approved it for submission: G Schwartzmann.

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