

Amusia following resection of a Heschl gyrus glioma

Case report

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✓ The incidence and character of neurological deficits following resection of glial neoplasms localized to the Heschl gyrus are currently unknown. In this series, the authors report the clinical presentation, management, and postoperative course of three patients with right hemisphere Heschl gyrus gliomas, one of whom developed difficulty with music production and comprehension postoperatively.

Resection of right hemisphere Heschl gyrus gliomas can result in deficits involving music comprehension. Preliminary evidence suggests that when these deficits occur, they may be transient in nature.

KEY WORDS • auditory cortex • brain neoplasm • Heschl gyrus • temporal lobe

PATIENTS with glial neoplasms localized to the primary auditory cortex or Heschl gyrus may present with seizures characterized by unique auditory or musical auras.^{8,13} The clinical presentation and treatment of patients with these rare lesions has not been well described. The only published discussion on this pathological entity was that by Silbergeld,¹³ who described two patients with gliomas localized to the Heschl gyrus of the dominant hemisphere, both of whom underwent complete resections of the lesions without auditory or language deficits postoperatively. He concluded that lesions confined to the Heschl gyrus are safe to remove from the dominant temporal lobe and that intraoperative language mapping was not required. In a subsequent study by Zatorre and Penhune,¹⁵ patients with epilepsy who had undergone unilateral temporal lobe excision that involved or encroached on the Heschl gyrus were tested for auditory dysfunction. These investigators reported that although resection or damage to the left hemisphere primary auditory cortex was not associated with auditory deficits, right hemisphere resections were commonly associated with auditory dysfunction, particularly worsened pitch discrimination. The incidence of auditory deficits following resection of right hemisphere Heschl gyrus gliomas is not known; there have been no cases reported in the literature thus far.

In this communication, we present the cases of three patients with surgically managed, nondominant, right hemisphere Heschl gyrus gliomas, one of whom had difficulty with music comprehension and production following surgery. The literature regarding clinical presentation, treatment, and possible complications for patients with Heschl gyrus gliomas is reviewed.

Summary of Cases

Case 1

This 28-year-old right-handed woman, a professional singer, presented with a generalized seizure that was preceded by an aura consisting of a loud sound of applause. An MR image revealed a probable low-grade nonenhancing glioma localized to the Heschl gyrus in the right hemisphere (Fig. 1A). A course of carbamazepine was prescribed and she never experienced another generalized seizure. She did, however, continue to have auditory auras consisting of short episodes of muffled sounds occurring once every few days. For tissue diagnosis and oncological cytoreduction and after induction of endotracheal anesthesia, a computer-assisted volumetric stereotactic resection of the patient's tumor was performed using a stereotactic system (COMPASS International Inc., Rochester, MN). Intraoperative transcranial motor and sensory evoked potential monitoring was performed, with no changes occurring during the resection. Postoperative MR imaging was used to confirm a gross-total resection (Fig. 2A). The pathological diagnosis was low-grade astrocytoma.

Immediately postoperatively the patient had markedly poor pitch discrimination and no melodic recognition (amusia). For example, she was unable to recognize or sing the tune for "Happy Birthday." Two weeks postoperatively she recovered her ability to produce scales with nearly accurate pitch. Her musical difficulties resolved completely by postoperative Week 3. She continued the course of carbamazepine and did not have any additional seizures or auras following tumor resection.

Case 2

This 27-year-old right-handed woman presented with auditory episodes characterized by a loud piercing beep,

Abbreviation used in this paper: MR = magnetic resonance.

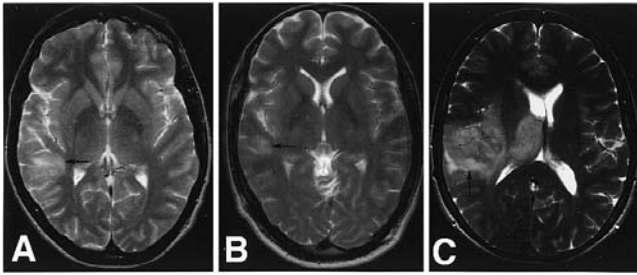


FIG. 1. Axial T₂-weighted MR images obtained in each of our three cases at the level of the Heschl gyrus. In all patients, the tumor was located in the right cerebral hemisphere (arrows). A: Case 1, with a 2 × 3-cm area of abnormal T₂ signal within the Heschl gyrus. B: Case 2, with an 8-mm area of abnormal T₂ signal within the gyrus. C: Case 3, with a T₂ signal abnormality centered on the Heschl gyrus and the planum temporale, with extension into the long gyri of the insula and ipsilateral thalamus. None of the tumors enhanced on T₁-weighted imaging after Gd injection (not shown).

similar to that made by the emergency broadcast system test sound. These episodes started suddenly, lasted a few minutes, and occurred every few weeks. An MR image was obtained that revealed a small, likely low-grade, nonenhancing glioma localized to the Heschl gyrus of the right hemisphere (Fig. 1B). A course of carbamazepine was initiated. For tissue diagnosis and oncological cytoreduction and after induction of endotracheal anesthesia, a computer-assisted volumetric stereotactic resection of her tumor was performed with the aid of the COMPASS stereotactic system. Intraoperative transcranial motor and sensory evoked potential monitoring was performed with no changes occurring during the resection. Postoperative MR imaging was used to confirm a gross-total resection (Fig. 2B). The pathological diagnosis was pleomorphic xanthoastrocytoma. She experienced no neurological or auditory deficits postoperatively. She continued taking carbamazepine and had no seizures during the follow-up period.

Case 3

This 46-year-old right-handed woman, a professor of

French, presented with generalized seizures that were preceded by a clanging noise in her head. An MR image revealed an infiltrating, nonenhancing glioma involving the Heschl gyrus of the right hemisphere with extension into the planum temporale, long gyri of the insula, and the right thalamus (Fig. 1C). After starting a course of phenobarbital, she never experienced another generalized seizure, yet she continued to have episodes of transient clanging, ringing, and rushing sounds on a weekly basis. Considering the diffuse nature of her tumor with involvement of the ipsilateral thalamus, a stereotactic biopsy was performed for tissue diagnosis. Pathological analysis revealed a low-grade fibrillary astrocytoma. Her antiepileptic medication was changed from phenobarbital to valproic acid and therapeutic levels were achieved. The auditory episodes subsequently abated. Conservative management with surveillance MR imaging was recommended.

Discussion

Topographic Anatomy of the Supratemporal Plane

Concealed within the sylvian fissure, the superior surface of the temporal lobe, or supratemporal plane, can be divided from anterior to posterior into three segments: the planum polare, the transverse temporal gyrus or gyri (Heschl gyrus), and the planum temporale (Fig. 3). Although a clear sulcal demarcation between the planum polare and Heschl gyrus is not often present on the supratemporal surface, a small sulcus (the acoustic sulcus) on the lateral surface of the superior temporal gyrus, which branches superiorly from the superior temporal sulcus, can be used to define the anterior extent of the Heschl gyrus. Posteriorly on the supratemporal plane, the transverse temporal sulcus is a reliable landmark that separates the Heschl gyrus from the planum temporale. Cytoarchitectural studies have shown that the primary auditory cortex usually covers the medial two thirds of the most anterior portion of the Heschl gyrus.⁶ Secondary auditory association areas include the planum temporale, superior temporal gyrus, parietal operculum, and long gyri of the insula.^{3,11}

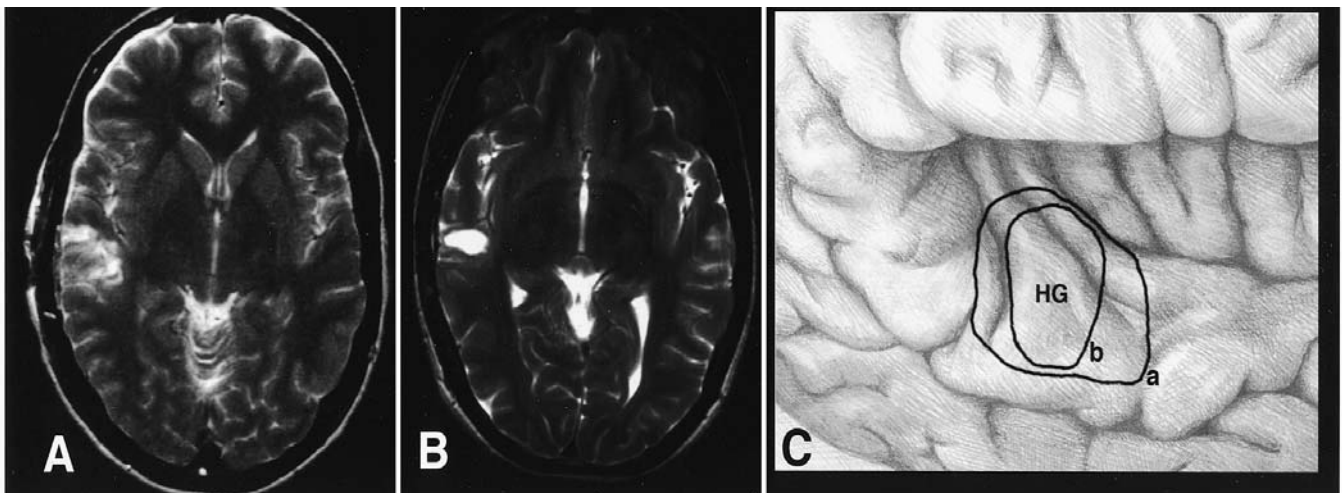


FIG. 2. Postoperative axial T₂-weighted MR images illustrating the extent of tumor resection in Case 1 (A) and Case 2 (B). Topographic drawing (C) of the primary auditory cortex with the extent of cortical resection for Case 1 (a) and Case 2 (b) schematically illustrated. HG = Heschl gyrus.

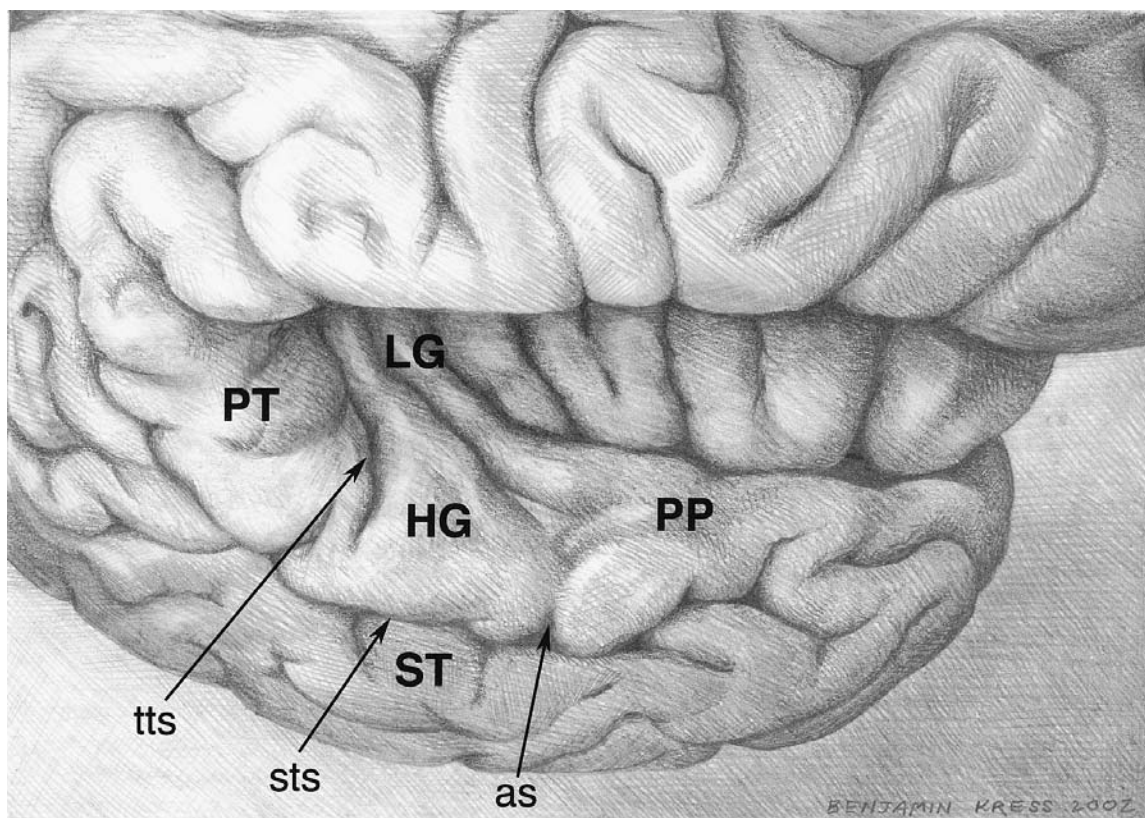


FIG. 3. An illustration of pertinent topographic anatomy of the supratemporal plane as depicted from a lateral view through the sylvian fissure of the right cerebral hemisphere. The Heschl gyrus is abnormally swollen with a glial neoplasm. The following structures can be identified: planum temporale (PT), Heschl gyri (HG), planum polare (PP), long gyrus of the insula (LG), superior temporal gyrus (ST), transverse temporal sulcus (tts), acoustic sulcus (as), and superior temporal sulcus (sts).

Anatomical and Functional Asymmetry of the Primary Auditory Cortex

One patient in our series had distorted perception of musical sounds that prevented her from singing for 3 weeks following resection of a right hemisphere Heschl gyrus glioma. Recently, there has been increasing evidence that the auditory cortices in each hemisphere are subspecialized, such that temporal resolution (important for speech discrimination) is better in the left auditory cortex, whereas spectral resolution (important for identifying melody) is better in the right auditory centers.¹⁴ Therefore, a deficit involving pitch discrimination or song production (which requires immediate auditory feedback) would not be surprising after damage to the right hemisphere auditory centers. Perry and coworkers¹⁰ reported asymmetrical increases in cerebral blood flow within the right Heschl gyrus,

measured by positron emission tomography while the patient sang, and concluded that its asymmetrical activation may be related to analyzing one's own voice for feedback-guided modulation. Additionally, it has been postulated that anatomical,^{1,4,12} cellular,⁴ and functional asymmetries² of the auditory cortices may have developed to optimize processing of the acoustic environment in both temporal (voice) and spectral (frequency, music) domains, thereby supporting the hemispheric auditory subspecialization theory.

Functional Plasticity and Response to Injury of the Primary Auditory Cortex

Although surgical manipulation causing dysfunction of nonresected secondary auditory cortex may be the most likely cause of our patient's transient deficit, functional plasticity could also have contributed to its resolution.

TABLE 1
Literature summary of surgically treated patients with Heschl gyrus gliomas

| Authors & Year | Age (yrs), Sex | Hemisphere | Aura/Seizure | Treatment | Outcome |
|------------------|----------------|------------|-------------------------|-----------|------------------|
| Silbergeld, 1997 | 41, M | lt | ringing, pure tone | resection | intact |
| | 34, F | lt | distortion of sound | resection | intact |
| present study | 28, F | rt | muffling of sound | resection | transient amusia |
| | 27, F | rt | ringing, rushing sound | resection | intact |
| | 46, F | rt | multiple piercing beeps | biopsy | intact |

Cortical representational plasticity is a well-documented phenomenon following neurological injury or after improved performance of perceptual and motor tasks. Both short- and long-term functional plasticity have been reported in the auditory cortex of healthy human volunteers following auditory frequency identification training demonstrated on functional MR imaging⁵ and magnetoencephalography studies.⁷ To the best of our knowledge our report is the first of right hemisphere, primary auditory cortex gliomas, of which two were completely resected, with one causing the patient to suffer a deficit in music perception. Functional plasticity of the auditory cortex or overall auditory processing may have restored the patient's ability to sing and comprehend music, which returned within 3 weeks of surgery. Silbergeld¹³ reported the resection of two left hemisphere Heschl gyrus gliomas by using awake intraoperative cortical stimulation mapping, with neither patient having a postoperative deficit. Although speculative, the left hemisphere auditory cortex may be more tolerant of surgical manipulation or removal because of its larger volume and greater number of pyramidal cells, more heavily myelinated axons, and greater interconnectivity when compared with the right hemisphere.^{4,9,14}

Clinical Presentation and Treatment of Patients With Heschl Gyrus Gliomas

Patients with Heschl gyrus gliomas have seizures characterized by auditory phenomena prior to (aura), during, or between epileptic episodes. In a total of five cases reported thus far, the hemisphere involved did not correlate with the pattern of presenting auditory symptoms (Table 1). Magnetic resonance images obtained in these patients reveal the location and extent of the tumors in the Heschl gyrus. Surgical removal of gliomas in this region is often indicated for tissue diagnosis and cytoreduction. Although Heschl gyrus lesions confined to the left hemisphere may be safe to remove without subsequent deficit, resection of right hemisphere lesions may cause difficulties with pitch discrimination and melodic recognition, as experienced by one of our patients. Considering the small number of cases reported in the literature thus far, the true incidence and clinical evolution of deficits following resection of Heschl gyrus gliomas is not known.

Conclusions

Patients with gliomas localized to the Heschl gyrus characteristically present with seizures having an auditory or musical component. Resection of right hemisphere Heschl gyrus gliomas can result in deficits involving music comprehension, which are likely to be transient. Further clinical experience is required to confirm these conclusions.

Disclaimer

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