Surgical treatment of partial epilepsy arising from the insular cortex

Report of two cases

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The role of the insula in partial epilepsy has been a source of interest and debate for several decades. Because of its multiple connections with the olfactory cortex, amygdala, entorhinal cortex, cingulate gyrus, and hippocampus, involvement of the insula in the genesis of limbic epilepsy is probable. Previous studies based on acute electrocorticography during temporal lobectomy, however, have indicated that insulectomy does not improve surgical results and may cause neurological deficits in patients with temporal lobe epilepsy who do not have structural lesions of the insula, even if the insular cortex is electrically abnormal. Because of its location deep within the sylvian fissure, scalp electroencephalography (EEG) is relatively insensitive to electrical activity in the insula and chronic invasive electrogaphic studies of the region have not been reported. We present two patients with partial epilepsy of insular origin who became seizure-free following insulectomy and excision of the intracranial mass. A review of the role of the insular cortex relative to partial epilepsy is also presented.

Case Reports

Case 1

This 13-year-old boy first developed seizures at 4 years of age. He had no previous neurological problems and his perinatal history was unremarkable. He experienced stereotyped seizures which started with a feeling of "butterflies in his throat." During this time he would place both hands over his upper chest. He would then begin rocking back and forth, occasionally using both arms and legs to accomplish this movement. The seizures lasted for 10 to 30 seconds. Postictally, he rapidly returned to his normal state and could remember words spoken to him during the ictal episodes. He would respond verbally to questions during ictus. He experienced several seizures per day.

Examination. Neurological examination was unremarkable, but magnetic resonance (MR) imaging revealed an intrinsic lesion involving the right insula and frontal operculum (Fig. 1 left). Scalp interictal EEG showed right frontotemporal slowing. At 11 years of age, the patient underwent an open biopsy of the frontal cortex for diagnosis. However, the pathology of the specimen showed only slight focal increase in astrocytes with minimal nuclear atypia. He was treated conservatively at that time, but several different anticonvulsant regimens produced poor seizure control.

At 13 years of age, the patient was readmitted for presurgical evaluation of his seizures. Scalp EEG during telemetry lateralized again to the right hemisphere. He underwent epicortical grid placement to document the cortical area involved during the ictal onset and to delineate the epileptogenic zone to be resected. Epicortical grids were placed over the frontal, temporal,
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Fig. 1. Magnetic resonance T1-weighted images of Case 1. Left: Preoperative image showing a high-intensity lesion in the right insula extending into the adjacent frontal operculum. Right: Postoperative image showing the resection cavity in the same area.

Fig. 2. Case 1. Left: Skull x-ray film showing the position of subdural electrodes for chronic epicortical grid recordings. These included a single eight-contact strip placed in the sylvian fissure so that the most distal electrodes recorded directly from the insular cortex. Right: Intraoperative photograph showing placement of the strip electrode in the sylvian fissure (the temporal lobe is at the top of the picture).

Fig. 3. Case 2. a and b: Preoperative magnetic resonance (MR) images, sagittal T1-weighted image (a) and coronal T1-weighted image (b), demonstrating an insular mass. c and d: Postoperative axial (c) and coronal (d) T1-weighted MR images showing the resection cavity in the posterior insula.

and parietal lobes. The sylvian fissure was also exposed and an eight-contact strip was placed to record directly from the insular cortex (Fig. 2). Chronic recording from the epicortical electrodes showed a regional ictal onset.

Operation. The patient subsequently underwent a right frontal opercular corticectomy and insulectomy. He tolerated the procedure well with no complications. Postoperatively, his neurological examination was unchanged. Pathological study of the specimen from this surgery revealed a low-grade pilocytic astrocytoma. At 24 months after surgery, he takes no anticonvulsant drugs and remains seizure-free. Postoperative MR images have shown no evidence of tumor recurrence (Fig. 1 right).

Case 2

This 24-year-old right-handed man suffered from complex partial seizures for 3 years after sustaining a minor head injury. The symptomatology included an aura involving a tingling sensation in his left arm and leg followed by a warm “flush” in his left arm and neck. He would then hyperventilate briefly, hypersalivate, display movements of his tongue, jaw, and mouth, and turn his head to the left. Next, he would perform complex activities with his arms and legs such as pulling at his bed sheets (crocodidmus) with occasional tonic posturing of the left arm and leg. He would not respond to verbal stimuli during the ictal event. Postictally he was confused and amnestic for the seizure. These seizures were unresponsive to medical therapy.

Examination. A gadolinium-enhancing solid mass localized to the right insular cortex was revealed on MR imaging (Fig. 3a and b). Routine EEG showed bilateral independent mesial temporal interictal spikes with more activity on the right side than the left. The patient underwent electroencephalographic telemetry which showed that the ictal onset was nonlocalizing. A detailed neuropsychological examination revealed a mild depression of cortical functions with a greater involvement of nondominant memory systems. Based on the EEG, it was believed that the patient was a poor candidate for a good surgical outcome with respect to seizure control and that the only indication for surgery would be for tumor resection in the event that the mass showed evidence of growth.

Operation. When the mass showed a progressive increase in size on serial MR imaging, the patient underwent a right frontotemporal craniotomy under general anesthesia with resection of the insular mass (Fig. 3c and d). After a standard frontotemporal craniotomy, the sylvian fissure was widely opened with the aid of the surgical microscope. Gentle retraction of the
frontal and temporal opercular cortices gave a broad exposure of the insula. At this point the sylvian fissure was filled with saline and intraoperative ultrasound imaging was used to outline the boundaries of the lesion. This tissue was then carefully removed by working between the branches of the middle cerebral artery until the resection was completed. The vessels in the sylvian fissure were soaked in papaverine prior to closure. The specimen was found to be a low-grade astrocytoma on pathological study. The patient did well postoperatively with no neurological deficits and at 24 months after resection is seizure-free without medication.

Discussion

The insula has been postulated to have a central role in linking sensory systems providing information about the external world with limbic and paralimbic systems involved with control of the internal milieu. The dorsal region of the insula has extensive connections with primary and secondary sensory neocortical structures, and neurons in this region respond to physiological, visual, somatosensory, and auditory stimuli. The ventral insula has extensive connections with limbic, paralimbic, and brain-stem autonomic centers. Neurons in this region respond to gustatory stimuli and stimulation of this area produces a decrease in blood pressure, inhibition of gastric peristalsis, and inhibition of respiration. Therefore, extensive involvement in limbic circuitry and function suggests a likely role for the insula in limbic seizures.

Physiological and anatomical studies on humans have concurred with many of the findings from animal experiments. Stimulation of the insula in awake patients undergoing temporal lobectomy elicits visceral motor, visceral sensory, and somatosensory responses. Intraoperative stimulation of the hippocampus produces evoked potentials in the insula. Visceral, gustatory, and somatosensory hallucinations have been emphasized in reports of seizures in patients with insular pathology. One report described generalized tonic-clonic seizures, which were abolished by resection of an insular lipoma. Reports of deficits following insular damage have given conflicting results. Although insulectomy sometimes produced hemiparesis ascribed to manipulation of the branches of the middle cerebral artery, Penfield and Faulk reported no detectable deficits referable to the surgical resection of the insula in patients undergoing temporal lobectomy for epilepsy. Studies of patients who have suffered infarcts of the insula, however, have disclosed global aphasia after dominant insular damage and mutism, neglect, apraxias, and bilateral opercular syndrome after non-dominant insular damage. It should be noted that pathological examination usually revealed damage to surrounding opercular cortex and corona radiata in these patients. In our two patients, there was no postoperative deficit that could be attributed to removal of the intrinsic insular lesions; however, both of these resections involved the nondominant hemisphere.

Our first patient reported a sensation of butterflies in the throat at the onset of his seizures. This is similar to previous reports of insular ictal phenomena involving visceral sensations in the throat, epigastrium, and abdomen. It is also consistent with the findings of Penfield and Faulk in patients with intraoperative insular stimulation. In our Case 1, chronic epicortical recording was useful in ruling out temporal lobe onset and in delineating the extent of epileptogenic cortex.

Our second patient had early onset of a tingling sensation in the contralateral leg. This was also seen in the series of Penfield and Faulk, where insular stimulation could produce sensations of varied character in the head, roof of the mouth, tongue, lips, arms, hand, trunk, legs, and toes. These were generally contralateral but could be bilateral or ipsilateral. Our Case 2 also demonstrated movements of the mouth and tongue, a motor effect elicited by intraoperative stimulation of the upper bank of the sylvian fissure. Although the symptomatology is not specific, a review of the reported cases shows that visceral sensory, somatosensory, and visceral motor phenomena occur commonly in seizures involving the insula. This raises the possibility that the insula is the source of auras when these symptoms persist after routine temporal lobectomy.

In a review of the experience of the Montreal Neurological Institute, Silfvenius, et al., concluded that in patients undergoing temporal lobectomy for temporal lobe epilepsy the addition of sulectomy did not improve surgical outcome even when the insula displayed interictal epileptiform activity on electrocorticography. It should be noted that patients with insular lesions were intentionally omitted from this study. The current report demonstrates the converse situation. In certain patients, an insular lesion can be the cause of partial seizures and its removal can result in resolution of the seizures even with the temporal lobe left undisturbed. Both of these patients had structural lesions of the insula. It is not known whether a similar situation exists in patients with no gross structural abnormalities. The fact that the insula is not monitored by standard chronic recording protocols suggests that this question will remain unanswered for some time.

In conclusion, this study expands our documentation of ictal phenomena which may be produced by a focus residing in the insula. It demonstrates that the region of the insula can be recorded chronically by a properly placed epicortical strip in the sylvian fissure. It shows that an insular lesion can be resected without removal of the overlying opercular cortices and without subsequent neurological deficits, and it demonstrates that partial seizures can be completely resolved with resection of the insular lesion and surrounding cortex.

References


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