Insular epilepsy masquerading as multifocal cortical epilepsy as proven by depth electrode

Case report

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The insular cortex is an uncommon epileptogenic location from which complex partial seizures may arise. Seizure activity in insular epilepsy may mimic temporal, parietal, or other cortical areas. Semiology, electroencephalography, and even surface electrocorticography recordings may falsely localize other cortical foci, leading to inaccurate diagnosis and treatment. The use of insular depth electrodes allows more precise localization of seizure foci. The authors describe the case of a young girl with seizures falsely localized to the cortex, with foci arising from the insula, as proven by depth electrode recordings. Resection of the insula yielded seizure control.

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Key Words • epilepsy • insula • depth electrode • topectomy • electrocorticography

Insular epilepsy is a rare form of complex partial seizures. Seizure spread from the insula may mimic typical temporal lobe epilepsy on EEG or surface ECoG recordings. Depth electrodes can monitor insular cortex. We describe a case of seizures localized to the insular cortex only after depth electrode recording. A 2-year-old girl suffered from intractable partial complex seizures; video EEG tracings suggested mesial temporal foci. Intraoperative ECoG showed interictal activity in the frontoparietal operculum, and topectomies were performed. Her seizures persisted, and implantation of depth electrodes defined the insula as the seizure focus. Radical insular resection eliminated seizures. Without insular depth electrodes, traditional localization strategies may be inaccurate in cases of seemingly classic neocortical epilepsy.

Case Report

History and Examination. This 2-year-old, right-handed girl experienced worsening intractable seizures beginning at 18 months of age. She had otherwise normal development and speech. Seizures were characterized as brief (20–40-second) episodes of staring, bilateral arm extension (more pronounced on the right), and leftward head version; they occurred between 20–30 times daily. Initial scalp EEG showed seizure activity at the C3 electrode, and video EEG 2 days prior to surgery indicated left temporoparietal seizure with a spread pattern. Magnetic resonance imaging revealed cortical dysplasia along the Sylvian region of the left temporal lobe extending into the insula (Fig. 1 left).

Operations. The patient underwent craniotomy and intraoperative surface ECoG with interictal activity over the frontal and parietal operculum and inferior frontoparietal cortex. Topectomies of these regions of dysplastic cortex were performed with the aid of stereotactic MR imaging guidance. A depth electrode placed parallel to the insula showed rare spike activity anteriorly, and this portion of the insula was resected. The total extent of the topectomy was approximately 5 cm from frontal operculum through the superior insula. The amygdala and mesial

Abbreviations used in this paper: ECoG = electrocorticography; EEG = electroencephalography.
temporal structures were spared. Immediately postoperatively, the patient’s seizures persisted unremittingly. She returned to surgery 4 days later, when surface grid and additional depth electrodes were placed along the long axis of the insula (Fig. 2 right). Using landmarks from the cortical surface, we placed the insular depth electrode parallel to the sylvian fissure. Telemetry showed 26 seizures with all onsets localized to the insular depth electrode and rapid spread to different areas of the cortical surface. The spread pattern varied across seizures with manifestations in lateral and inferior temporal, posterior parietal, and supraorbital surface electrodes (Fig. 2).

After 3 days of monitoring, a radical insular topectomy was then performed with ECoG guidance. Spike-and-wave activity was noted in the superior temporal gyrus and frontal operculum. The opercular resection was extended to the margin of the sylvian fissure. Insular vessels from the middle cerebral artery were identified and spared, and the topectomy continued to the previous resection bed in the anterior insular cortex. Cortical motor mapping was applied; however, responses were absent, limiting to 1 cm the depth of anterior insular resection. The posterior aspect of the insula was also noted to be spongy in appearance and texture, consistent with dysplasia; this was extensively resected to normal tissue (Fig. 3). Following these topectomies, intraoperative ECoG showed no epileptiform activity from the insular cortex and only rare spike-and-wave activity in the occipital cortex.

**Postoperative Course.** Her recovery included a 22-month seizure-free period, followed by occasional complex partial seizures (2–3/month) with right frontal interictal abnormalities on video EEG. Her seizures are controlled with a single antiepileptic agent, and overall seizure frequency was markedly reduced compared with preoperatively, without episodes of status epilepticus. She continues to develop normally and is doing well in school.

**Discussion**

Since first described by Penfield and Faulk, seizures arising from the insular cortex have become increasingly recognized, although they are rare. Seizures originating...
Depth electrodes for insular epilepsy

in this area can produce central symptoms (nausea, dysgeusia, emotion, and somesthesis), and spread to adjacent surface cortex can produce motor symptoms. Conversely, the central symptoms of mesial temporal lobe epilepsy have been shown to arise from the spread of seizure activity to the adjacent insula. Studies of patients with documented insular epileptic foci have shown that conventional video-EEG monitoring cannot be relied on to differentiate between temporal and insular discharges.

Invasive monitoring, such as grid ECoG and subdural strip electrodes, has been used to successfully localize seizure foci in intractable epilepsy when conventional EEG is unrevealing. However, seizures arising from deeper structures (such as the insula) can spread to the cortical surface. In such cases, direct recording by depth electrodes aids in localization. If the insula is not sampled, rapid spread to neocortex can be unpredictable.

This case demonstrates how the spread of epileptogenic activity from the insula to adjacent cortex may be falsely localized to the cortical surface by scalp EEG and even ECoG, leading to persistence of seizures after topectomy. Lobar corticography alone would have incorrectly localized these seizures and incorrectly classified the seizures as multifocal. Only by interpretation of depth electrode data can insular epilepsy be localized and treated. Insular electrode implantation is a safe method of determining primary versus secondary insular involvement prior to surgery. Previous work has shown that, although combining conventional temporal lobectomy with insulectomy did not reduce seizures in patients with temporal lobe epilepsy, some patients in whom the previous temporal lobectomy failed experienced a significant seizure reduction after undergoing reoperation and insulectomy. That study, done without depth electrodes, implied that patients ultimately shown to have insular onset may appear to have temporal lobe origin. Our case underscores the challenges of localizing an insular focus even with neocortical ECoG.

Disclosure

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

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References


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