INNOVATIVE COLLECTIONS

COMPLEX CASE STUDY

Asystole Induced by Recurrence of Adenoid Cystic Carcinoma: A Case Report of Ictal Asystole and Discussion of Management Strategies

JUYONG BRIAN KIM, MD, MPH and DAVID A. CESARIO, MD, PhD

Division of Cardiology, Department of Medicine, David Geffen School of Medicine at University of California, Los Angeles, CA
Director of Cardiac Electrophysiology, Cardiovascular Keck School of Medicine, University of Southern California

ABSTRACT. Asystole and profound bradycardia related to seizure activity are rare; however, these entities appear to be gaining prominence as a possible etiology of sudden unexpected death in epilepsy (SUDEP). We report a case of prolonged asystole induced by seizure activity from a tumorigenic focus.

KEYWORDS. asystole, seizure, SUDEP, adenoid cystic carcinoma, pacemaker.

Introduction

A potential mechanism of sudden unexpected death in epilepsy (SUDEP) is epilepsy-induced cardiac bradyarrhythmia or asystole. Most frequently, epilepsy is associated with sinus tachycardia; however, an increasing number of bradyarrhythmic events have been reported. The optimal management and treatment of patients who present with these events has not yet been established. We report a case of a 67-year-old woman with asystole induced by seizure activity from a tumorigenic focus, and discuss the related literature and review the current management strategies.

Case report

A 67-year-old right-handed woman presented to the emergency department with a 1-week history of recurrent daily episodes of syncope. Each episode was preceded by a metallic olfactory sensation, followed by a transient loss of consciousness for 1–2 min. The first episode resulted in a motor vehicle accident; two subsequent episodes were witnessed by her husband, where she was noted to have bilateral asymmetric jerking motions of her upper extremities, and a staring gaze associated with her loss of consciousness. There was no urinary or fecal incontinence. The patient’s past medical history includes, at age of 65, resection of adenoid cystic carcinoma of the right maxillary sinus engulfing the infra-orbital nerve with clean margins, followed by 6 weeks of intensity-modulated radiation therapy. Her other history includes cerebral aneurysm status post coiling, as well as partial resection of thyroid due to cancer, migraine headaches, and a history of anxiety. She denied having any previous episodes of syncope or seizure-like activities. The patient has residual diplopia and right-sided facial numbness that has not worsened over time.

In the emergency room (ER), the patient was initially found to be in normal sinus rhythm with stable vital signs. The patient was subsequently witnessed to have a laterally deviated gaze with mild jerking movements of her upper extremities and on telemetry developed profound bradycardia followed by 19 s of asystole. The cardiac rhythm then returned to normal sinus rhythm with subsequent recovery of consciousness and neurologic function (Figure 1). Blood pressure was within normal range upon recovery. The patient had a short period of postictal confusion lasting several minutes after her seizure. Transcutaneous pacing pads were placed

The Journal of Innovations in Cardiac Rhythm Management, April 2011
Figure 1: An event recording from a continuous cardiac monitor shows severe bradycardia followed by 19 s and 5 s of asystole separated by a junctional beat and followed by a run of junctional beats. Return to normal sinus rhythm is seen at the end of the strip.
A case report of ictal-asystole and discussion of management strategies

on the patient, and she was empirically started on a loading dose of dilantin, and maintained on antiepileptic treatment during her hospitalization. Imaging studies including magnetic resonance imaging (MRI) of the brain and a positron emission tomography (PET) scan revealed focal fluorodeoxyglucose uptake in the region of the inferomedial right temporal lobe compatible with recurrence of the adenoid cystic carcinoma with extension into the mesiotemporal lobe (Figure 2). Once a complete set of magnetic resonance imaging studies were obtained in consultation with the neurological, neurosurgical, and radiation-oncology services, a permanent dual-chamber (DDD mode) pacemaker was placed, and the patient was discharged on antiepileptic medications. One year following the event, this patient remains free of syncopal episodes, although she reports having two episodes of olfactory aura. An electroencephalogram performed after the initial event did not show any sign of epilepsy. The patient continues to do well to date.

Discussion

Nearly one-fifth of all deaths in patients with epilepsy are SUDEP. The incidence of SUDEP is estimated to be at 1.21/1000 patient-years. However, there is no estimation of what proportion of SUDEP is attributable to ictal asystole. Ictal bradyarrhythmias are thought to be rare, as there have only been approximately 80 cases reported in the literature to date; however, the true incidence is likely underestimated given the difficulty of documenting bradycardic/asystolic activity and epileptic activity concurrently. Rugg-Gunn et al. noted that 7 out of 19 patients with refractory partial seizures in their study had ictal bradycardia on implantable loop recorder monitoring, suggesting a higher incidence in patients with refractory epilepsy. Unfortunately, most cases occur in the absence of video electroencephalogram/electrocardiogram (EEG/ECG) monitoring. However, in this patient we were fortunate to have a witnessed seizure episode while the patient was on a cardiac monitor and thus incidentally noted the prolonged asystolic arrest.

Recognition of ictal asystole is important for those evaluating a patient for epilepsy, syncope, and other episodic disorders, as it has important therapeutic implications. It is easy to consider a primary cardiogenic diagnosis over a neurologic diagnosis or vice versa, when, in fact, the two systems may manifest together. A retrospective analysis of video-EEG/ECG monitoring over 14 years by Gearing et al. at the Mayo Clinic found 13 patients with ictal bradycardia or asystole. The study proposed that the presence of loss of muscle tone or bilateral asymmetric jerky limb movements during a seizure suggests the possibility of ictal asystole. This case supports this notion in that the patient had syncopal episodes presenting with visual or olfactory auras, and bilateral jerking movements of upper limbs that spontaneously resolved. However, there are similarities in the clinical features of non-epileptic syncope and ictal asystole that suggest that the above findings are due to cerebral hypoperfusion secondary to asystole rather than stimulation of the cortical and subcortical regions by the seizure activity.

The duration of cardiac asystole is usually short, averaging 5–10 s in the previously reported cases, with one exception when 30 s of asystole was observed during a seizure. The patient in this case had 19 s of asystole, which suggests that prolonged episodes of asystole are possible increasing the risk of SUDEP. It is likely that prolonged asystolic episodes have not been captured, as patients are less likely to survive these episodes.

There are currently no clear risk factors for ictal asystole other than refractory partial seizures, and patients commonly do not have any previous history of cardiac disease or other cardiovascular risk factors. In fact, the mechanism of induction of bradyarrhythmia or asystole is poorly understood. A proposed mechanism is that seizure activity induces a surge in vagal tone, leading to temporary bradyarrhythmias. However, no consistent pattern has been found regarding the location of the seizure activity and the type of arrhythmia. In our patient, an epileptic focus in the right temporal lobe appeared to trigger a neurocardiogenic stimulus that may have resulted in asystole. Based on the reports to date, the most frequently reported culprit location for epileptic foci resulting in bradyarrhythmias appears to be temporal lobe partial seizures. Additionally, Oppenheimer et al. demonstrated that stimulation of the insular cortex can produce heart block. It is noteworthy that several cases of asystole have been reported during electroconvulsive therapy, which may provide an opportunity for further studies of the mechanism of this phenomenon.

This case also illustrates the importance of follow-up monitoring of treated intracranial tumors. In the

Figure 2: A cross-sectional T2-weighted magnetic resonance image shows hyperintensity within the right temporal lobe (white arrow), representing recurrence of adenoid cystic tumor in this patient.
described patient, tumor recurrence first presented in the form of a life-threatening cardiac event. Although the current practice recommends against prophylactic use of antiepileptic drugs in patients with brain tumors, early initiation of antiepileptic medication even in the absence of any seizure activity may need to be considered in select patients. In fact, any patient with a current intracranial process should be considered for antiepileptic medications based on the location and size of the process, given that the first event may be fatal. However, the selection of patients who would require long-term antiepileptic medications among patients without known seizure activity can be difficult. In these patients, early video-EEG/ECG monitoring should be considered for further workup.

It should also be noted that the incidence of partial seizures significantly increases in the elderly population. Prophylactic treatment with antiepileptic medications in all patients with intracranial mass or history of partial seizure activity is obviously controversial and the cost-benefit analysis for such treatment strategies is not known. Future studies including large population case-control studies would help answer this question. However, it may be reasonable to have a lower threshold for the placement of implantable loop recorders in patients with a history of partial seizures and syncope to diagnose occult bradyarrhythmias and consider permanent pacemaker implantation in appropriate individuals.

Although the diagnosis of ictal asystole requires simultaneous EEG and ECG records, initiation of antiepileptics and consideration for pacemaker placement should not be delayed if clinical suspicion is high, as video-EEG/ECG testing requires inpatient hospital stays incurring high costs and may not yield a definitive diagnosis as the timing of subsequent events cannot be predicted. Additionally, a sufficient number of seizure episodes needs to be recorded in order to rule out ictal asystole, as not all epileptic episodes lead to bradyarrhythmias.

Finally, the initial management of patients presenting with probable ictal-arrhythmic events should include MRI of the brain to evaluate for an anatomical foci for their seizures. These imaging studies should precede the placement of a pacemaker for prophylactic treatment of neurocardiogenic syncope or SUDEP. Any space-occupying lesion that may be an epileptic source should be thoroughly evaluated prior to pacemaker implantation in collaboration with specialists, including neurologists, neuroradiologists, neurosurgeons, radiation oncologists, and cardiologists.

Overall, this case illustrates several interesting points about the importance of close monitoring of patients with recurrent syncopal episodes, even if they are thought to be related to seizure activity. In such patients treating physicians should have a heightened suspicion for ictal asystole as a cause of syncope and should consider additional workup, including holter monitoring, event monitoring, implantable loop recorders, video electroencephalogram–electrocardiogram testing, and even permanent pacemaker placement in select patients.

References