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Case Report

Intractable Yawning as a Predominant Symptom of Temporal Lobe Ganglioglioma: Case Report and Review of Literature

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Abstract

Yawning, a physiologic reflex exhibited by vertebrates, is seldom noticed as a symptom of a disease. Not too often is a patient aware of it as a symptom, unless it is of such a distressing nature to seek attention. In this situation, to distinguish between normal and abnormal behavior would pose a diagnostic dilemma for the attending physician. Intractable yawning has been a presenting symptom of many pathologic states such as stroke, epilepsy, and migraine. Literature is sparse regarding intractable yawning caused by tumors of the brain. Most of the time, the etiology cited is the infratentorial location of these tumors causing compression of the brainstem and the centers responsible for yawning. Intractable yawning as a predominant symptom of supratentorial tumor is rare. We present a case of an 18-year-old girl who presented with abnormal yawning. On evaluation, magnetic resonance imaging revealed a tumor in the posterior part of the inferior temporal gyrus. There was no significant compression of the brainstem structures to suggest this as a cause for her symptom. She underwent a craniotomy and total excision of lesion. Postoperatively, her symptoms improved and her salvos of yawns ceased. The histopathological examination revealed a ganglioglioma of the temporal lobe. The present case is unique as it is the only case reported in the literature of a supratentorial tumor causing abnormal yawning.

Keywords: *Ganglioglioma, intractable yawning, temporal lobe*

Introduction

Yawning is a physiologic reflex controlled by a complex set of neurons situated in and around the brainstem. There are many theories to the origin of this natural phenomenon described in the literature. Common to them, the paraventricular nucleus describes special mention as it is cited to be the integration center of this multi-phased reflex. Often this is accompanied by pandiculations, a phenomenon whereby it is associated with stretching and raising of arms. Rarely, this reflex is unchecked in individuals in various diseased as well as iatrogenic states. When this happens, the patients throw salvo of yawns otherwise known as "chasms." There are diverse definitions of what constitutes abnormal yawning. Many authors have described it ranging from 2/10 to 30/10 min. Regardless of the number, it leaves one with no doubt as to what "abnormal" is, when the patients seek attention for it. Among few reports that cite a brain tumor as the etiology, most all of them have been due to tumors in the infratentorial compartment compressing the brain stem. We encountered a supratentorial tumor in the temporal lobe as the cause of intractable yawning.

Case Report

An 18-year-old girl presented with a history of the absence of awareness which lasted for 1–2 min followed by bouts of abnormal yawning of 24 months duration. She used to yawn at least 20–30 times in succession after which she would have mild headache followed by drowsiness. She had at least 10–12 such episodes at irregular intervals during a day. She was treated by a local physician who initiated her treatment with carbamazepine. She was not responding well to it when clonazepam was added as the second drug. There was a change in her semiology of symptoms where by her symptoms of loss of awareness subsided and she was left only with these bouts of intractable yawning. She had a lot of weight gain as an adverse effect of carbamazepine when she was referred to us. She underwent magnetic resonance imaging (MRI) which showed a well-circumscribed lesion of

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about 2 cm × 2 cm in the posterior part of the right inferior temporal gyrus [Figure 1a-d]. The tumor was partly cystic with an enhancing mural nodule. There was no significant mass effect or any midline shift. There were no areas of blooming on gradient echo. There was no hydrocephalus. An interictal electroencephalography (EEG) was taken which revealed focal slowing over the right temporal region.

She had no previous history of seizures. There was no history of sleep deprivation. On examination, she was fully conscious and oriented. Her Epworth sleep scale score was 2/10. She had no history of sleep apnea. She had no cranial nerve deficit. There was no weakness involving any of the limbs. She had only yawning as the complaint which was witnessed during the entire hospital stay. She underwent right temporo-occipital craniotomy and total excision of the tumor. The tumor was well-circumscribed with well-defined arachnoid plains. It was moderately vascular and was excised totally. A histopathological examination showed her lesion to be ganglioglioma [Figure 2a and b]. Postoperatively, her frequency of yawning decreased. We were able to reduce the dosing of her anticonvulsants. She was weaned off from each gradually by the end of one year and by that time she was totally free of her symptoms. MRI was repeated after 5 years and there was no recurrence of the lesion [Figure 3a-d]. She has been on our follow-up for last 5 years, and she has been symptom-free until now.

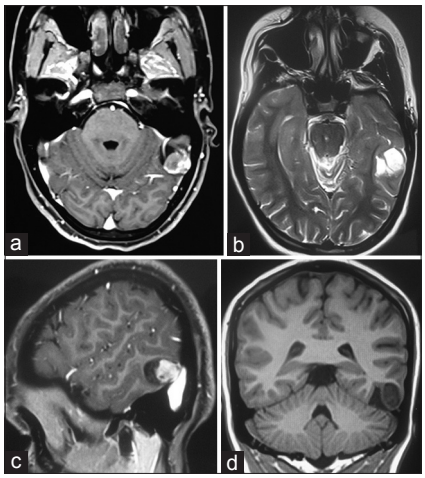

Figure 1: (a) Axial T1-weighted with gadolinium enhancement. (b) Axial T2-weighted. (c) Sagittal T1-weighted with gadolinium enhancement. (d) Coronal T1-weighted with FLAIR. Preoperative images showing a lesion of size about 2 cm × 2 cm involving the posterior part of the right inferior temporal cortex

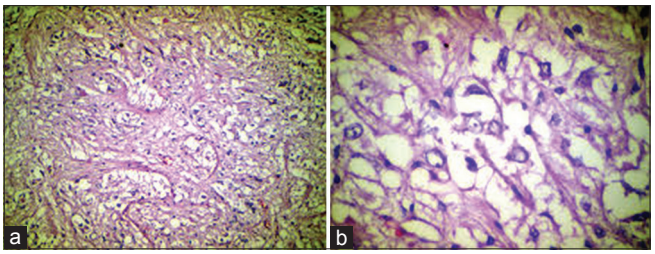

Figure 2: (a) Photomicrograph - Low power view of the tumor showing a biphasic pattern of abnormal clustered neurons and fibrillary astrocytoma components. (b) Photomicrograph - High power view showing the abnormal ganglion cells

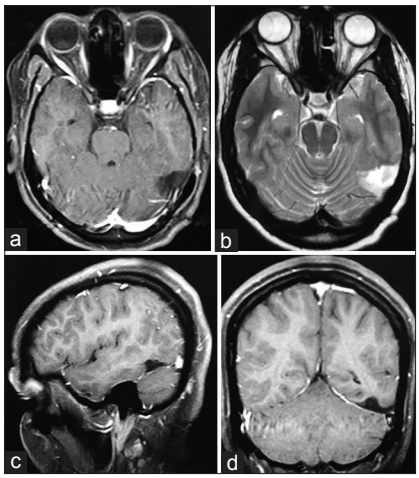

Figure 3: (a) Axial T1-weighted with gadolinium contrast. (b) Axial T2-weighted. (c) Sagittal T1-weighted with gadolinium contrast. (d) Coronal T1-weighted with gadolinium contrast. Postoperative images 5 years after surgery showing no recurrence of the lesion

Discussion

Yawning as a sequelae to tumors of the brain is rare. Almost all of them are reported to be due to infratentorial tumors.[1,2] Although the occurrence of this phenomenon with supratentorial pathology such as stroke is common[3,4] it has never been reported with tumors in that region. A thorough search on PubMed, with terms intractable yawning, brain tumors, and supratentorial did not reveal any results. Hence, we believe this is the first case report of intractable yawning as the cause of a supratentorial tumor.

The mechanisms underlying the occurrence of yawning are arcane. There are at least three neurogenic pathways that have been identified in the induction and control of yawning. All of them cite, the paraventricular nucleus of the hypothalamus and their connections as the prime integration unit of this circuit.[4] Yawning has been described occurring as a preictal, ictal as well as postictal phenomenon. When occurring in the preictal phase, it has a localizing value as it indicates its origin in the nondominant temporal lobe.[5] Our patient had a tumor involving the nondominant temporal lobe. Jasper and Penfield were the first to describe their patient with diencephalic epilepsy in which their patient had yawning and other dysautonomias in addition to seizures. Our patient had only loss of awareness other than intractable yawning. We believe that this was itself an ictal seizure manifestation. Unfortunately, due to the lack of video EEG or intraoperative electrocorticography in our institution, this was not done. Ictal yawning has been reported before although rare.[6,7] Most of the pathology responsible for abnormal yawning reported so far have been located in close

proximity to the brainstem.[1,2,8] In our patient, the lesion was located in the posterior part of the inferior temporal lobe. There was no mass effect on the brainstem to attribute it to the cause of yawning. The histology of the lesion in our patient was ganglioglioma, a glioneuronal tumor which is a leading cause of long-term epilepsy associated tumor, also known as LEAT. The intrinsic nature of cells of ganglioglioma predisposes to epilepsy. Glioneuronal tumors are composed of peculiar cellular components with hyperexcitable neurons, functionally integrated into excitatory circuitries, and neurochemical characteristics that can be relevant for epileptogenesis.[9]

Abnormal yawning also has been reported in supratentorial nonorganic lesions such as stroke. The etiology cited in such instances have been that supratentorial lesions release their inhibitory effect on the yawning centers of brainstem.[9] The fact that her symptoms were mitigated immediate postoperatively denotes that it was infact, the focus of seizure which manifested as intractable yawning in our patient.

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Conflicts of interest

There are no conflicts of interest.

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