CASE REPORT



A case of syncopal convulsions triggered by glossopharyngeal neuralgia

Glossofarengeal nevraljinin tetiklediği senkopal konvülziyon olgusu

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Summary

Syncopal convulsions and epileptic seizures are clinically hard to distinguish and differ in terms of treatment approaches. It is important to consider the cardiac arrhythmias that impair cerebral perfusion in the differential diagnosis of antiepileptic treatment-resistant convulsions. Here, we offer a 72-year-old male patient glossopharyngeal neuralgia (GN) after swallowing associated with recurrent episodes of syncopal convulsions. The patient was successfully treated with temporary pacemaker and carbamazepine. This phenomenon is noteworthy in terms of both asystole triggered by GN and syncopal convulsions which are rare in the differential diagnosis of epileptic seizures.

Keywords: Asystole; glossopharyngeal neuralgia; syncopal convulsion.

Öz

Senkopal konvülziyonların ve epileptik nöbetlerin klinik olarak ayırtedilmesi zordur ve tedavi yaklaşımları farklılık gösterir. Antiepileptik tedaviye dirençli konvülziyonların ayırıcı tanısında serebral perfüzyonu bozan kardiyak aritmileri düşünmek önemlidir. Bu yazıda, yutkunma ile ilişkili tekrarlayan senkopal konvülsiyon atakları olan 72 yaşındaki glossofarengeal nevraljili erkek hasta sunuldu. Hasta geçici kalp pili ve karbamazepin ile başarılı bir şekilde tedavi edildi. Bu olgu, epileptik nöbetlerin ayırıcı tanısında nadir görülen glossopharengial nevralji ve buna bağlı asistolinin tetiklediği senkopal konvülziyonlara dikkat çekmektedir.

Anahtar sözcükler: Asistoli; glossofarengial nevralji; senkopal konvülziyon.

Introduction

Glossopharyngeal neuralgia (GN) is a disease characterized by paroxysmal sharp, stabbing, shooting, lancinating flashes of excruciating to agonizing "electrical shock-like" or "needle-like" pain on posterior region of the tongue, tonsils, oropharynx, larynx, auditory canal, middle ear, angle of the mandible, and sometimes the retromolar region.^{[11} Transient, self-limited interruptions of cardiac output result in generalized cerebral ischemia, a condition that is termed syncope when it results in a loss of consciousness.^{[21} The clinical spectrum of abnormalities that occur with generalized cerebral hypoperfusion is broad, ranging from non-specific dizziness to a variety of sensory disturbances, including paresthesias and alterations of vision, to loss of consciousness, sometimes with convulsive features.^[3] The frequency of cardiac syncope in GN is 2–20%.^[4]

Case Report

A 72-year-old male patient with no known medical history was admitted to the emergency department with a complaint of seizures lasting about 10–15 s while the eyes remained open and fixed and the whole body stiffened numerous times a day for the past week. Routine blood tests (complete blood count, glucose, AST, ALT, urea, creatinine, sodium, potassium, chlorine, and calcium) and cranial computed

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tomography showed no pathological findings. Electrocardiogram (ECG) demonstrated a sinus rhythm of 75 beats/min. The patient was admitted to our clinic for follow-up and treatment with an early diagnosis of epilepsy. Cranial magnetic resonance imaging and electroencephalography (EEG) were performed for the etiology of epileptic seizures. No pathological findings were detected in the cranial magnetic resonance imaging and EEG scans. In our clinical follow-ups, the seizure frequency was found to be 6-10 times a day and the seizures were determined to be generalized tonic-clonic seizures. When the medical history of the patient with resistant seizures was detailed, it was learned that the seizure started pain like an electric shock in the throat and that this pain occurred after swallowing. Cardiac monitoring was performed in the patient who was diagnosed as having epileptic seizures after GN as this patient may have been suffering from reflex asystole (Fig. 1). After swallowing, the patient developed asystole followed by a 10 s syncopal convulsion. Postictal confusion, urinary incontinence, and tongue biting were not observed. Carbamazepine 600 mg/day was administered for GN and control of seizures to patients. However, the seizures could not be controlled with carbamazepine therapy. A cardiology consultation was requested and the patient was fitted with a temporary pacemaker. There were no epileptic seizures after temporary pacemaker implantation. No pain and seizure were determined with 1200 mg carbamazepine therapy without pacemaker in the control examination performed 1 week later.

Discussion

GN may be accompanied by recurrent episodic hypotension, bradycardia, syncope, seizures, or even cardiac arrest.^[1] Eating and speaking trigger neuralgia.^[5] Changes in blood pressure and cardiac rhythm, and bradycardia were induced by stimulation of the larynx.^[6] Syncope in GN related to neuralgic pain is most likely caused by activation of the dorsal motor nucleus of the vagus nerve by abnormally enhanced input from afferent or ischemic lesions of the glossopharyngeal nerve. The reflex arrhythmia could be explained from the fact that afferent nerve impulses from the glossopharyngeal nerve may reach the tractus solitarius of the brainstem and through collateral fibers reach the dorsal motor nucleus of the vagus nerve. Activation of this abnormal loop dur-



Figure 1. Cardiac monitorization image of the patient of syncopal convulsions triggered by glossopharyngeal neuralgia. (a–c) Bradycardia and then asystole occurred after swallowing. (d–f) Artifact due to convulsion was observed in monitor. (g and h) Convulsions stopped when the heart beat returned to sinus rhythm.

ing severe neuralgic pain would be responsible for bradycardia/asystole, with cerebral hypoperfusion, slowing of EEG activity, syncope, and convulsions in proportion to the duration of asystole.^[7] This phenomenon is noteworthy in terms of both asystole triggered by GN and syncopal convulsions which are rare in the differential diagnosis of epileptic seizures.

In a videometric analysis of syncope induced in 42 of 59 healthy volunteers, lasting 12.1±4.4 s, 90% (38 volunteers) myoclonic activity occurred. Head turns, oral automatisms, and righting movements were prominent in these seizures. Initial upward deviation was common and eyes remained open in 76% of cases. About 60% reported visual hallucinations, and 36% were accompanied by auditory hallucinations and understandable speech was never observed. ^[8] The association of GN with cardiac syncope has rarely been described in the literature.^[9] Focal neurological symptoms are rare with cardiac arrhythmias. In a study of 290 patients who received pacemakers, only 4 (1.4%) had transient focal neurological symptoms and signs. Syncope occurred in 167 (57.6%) of 290 patients. Generalized seizures occurred in 26 patients (8.9%); about half of these had a preceding "warning" of feeling faint, and about half had their seizure abruptly without warning. Focal motor or sensory seizures did not occur in any patient.^[10] Cardiac arrhythmias are particularly common in temporal lobe seizures. Seizure-related sinus tachycardia may be detected from the onset of temporal and extratemporal seizures. Bradyarrhythmias such as bradycardia, sinus arrest, atrioventricular block, and asystole are rare seizure-related cardiac dysrhythmias. In these cases, interictal electroencephalography (EEG) and electrocardiographydata were typically within normal range.^[11] Asystole was detected in our case, and our interictal EEG data were normal.

In a joint study of cardiologists and neurologists, were presented a simple point score of diagnostic criteria that distinguishes syncope from seizures with high accuracy. Some questions about symptoms of seizure and syncope were used in scoring. The symptoms which are cut tongue, deja vu, limb jerking, and postictal confusion all contributed to the diagnosis of seizures. Symptoms of syncope such as prodromal diaphoresis and palpitations, or provocation by prolonged sitting or standing, often have needed to be absent to diagnose a seizure.^[12]

The medical literature supports the use of carbamazepine in the management of idiopathic neuralgia at treatment.^[13] Temporary pacemaker can be used to treat the reflex cardiac syncope until therapeutic levels of carbamazepine.^[14]

Conclusion

As it can be seen, syncopal convulsions, a rare entity in GN and epileptic seizures, are clinically hard to distinguish and differ in terms of treatment approaches. It is important to consider the cardiac arrhythmias that impair cerebral perfusion in the differential diagnosis of antiepileptic treatment-resistant convulsions. Our case draws attention to this issue.

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying image.

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References

- 1. Blumenfeld A, Nikolskaya G. Glossopharyngeal neuralgia. Curr Pain Headache Rep 2013;17(7):343. [CrossRef]
- 2. Kapoor WN. Syncope. N Engl J Med 2000;343:1856. [CrossRef]
- 3. Lin JT, Ziegler DK, Lai CW, Bayer W. Convulsive syncope in blood donors. Ann Neurol 1982;11(5):525–8. [CrossRef]
- Rushton JG, Stevens JC, Miller RH. Glossopharyngeal (vagoglossopharyngeal) neuralgia: A study of 217 cases. Arch Neurol 1981;38(4):201–5. [CrossRef]
- Bradley WG, Daroff RB, Fenichel GM, Jankovic J. Otonom sinir sistemi hastalıkları. In: Tan E, Ozdamar SE, editors. Neurology in Clinical Practice. Ankara: Veri Medikal Yayıncılık; 2008. p. 2376.
- Barbash GI, Keren G, Korczyn AD, Sharpless NS, Chayen M, Copperman Y, et al. Mechanisms of syncope in glossopharyngeal neuralgia. Clin Neurophysiol 1986;63(3):231– 5. [CrossRef]
- Den Hartog AW, Jansen E, Kal JE, Duyndam D, Visser J, Van Den Munckhof P, et al. Recurrent syncope due to glossopharyngeal neuralgia. Heart Rhythm Case Rep 2017;3(1):73–7. [CrossRef]
- Lempert T, Bauer M, Schmidth D. Syncope: A videometric analysis of 56 episodes of transient cerebral hypoxia. Ann Neurol 1994;36(2):233–7. [CrossRef]
- 9. Burfield L, Ahmad F, Adams J. Glossopharyngeal neuralgia associated with cardiac syncope. BMC Case Rep 2016;2016:bcr2015214104. [CrossRef]
- Reed RL, Siekert RG, Merideth J. Rarity of transient focal cerebral ischemia in cardiac dysrhythmia. JAMA 1973;223:893–95. [CrossRef]
- 11. Bora I, Yeni SN, Gürses C. Epilepsi. 1st ed. İstanbul: Nobel Kitabevleri; 2008.
- 12. Sheldon R, Rose S, Ritchie D, Connolly SJ, Koshman ML, Lee MA, et al. Historical criteria that distinguish syncope from seizures. J Am Coll Cardiol 2002;40:142–8. [CrossRef]
- 13. Johnston RT, Redding VJ. Glossopharyngeal neuralgia associated with cardiac syncope: Long term treatment with permanent pacing and carbamazepine. Br Heart J 1990;64(6):403–5. [CrossRef]
- 14. Khero BA, Mullins CB. Cardiac syncope due to glossopharyngeal neuralgia treatment with transvenous pacemaker. Arch Intern Med 1971;128(5):806–8. [CrossRef]