e-ISSN:2147-2181

causapedia
hakemli olgu dergisi

Bipolar Disorder: A Result of Tuberous Sclerosis Complex?

Bipolar Bozukluk, Tuberoz Skleroz Kompleksinin Bir Sonucu mu?
Bipolar Bozukluk Tanısı ile Geç Yaşta Prezente Olan Nadir Bir Olgu
Nöroloji

Başvuru: 18.09.2014
Kabul: 07.11.2014
Yayın: 24.11.2014

Meltem Duraklı Ulukök[1], Hasan Meral[2], Tuba Özcan[3], Gülçin Yücel[2]

[1] Cankaya Medical Center
[2] Çanakkale Anatolian Hospital
[3] Ordu Üniversitesi Tıp Fakültesi

Özet

Tuberoskleroz kompleksi (TSK), otistik bozukluk sprektrumu, hiperkinetik bozukluklar, depresyon ve anksiyete bozukluğu gibi önemli psikiyatrik tablolarla komorbidite göstermektedir. Ancak bipolar bozukluk ile birliktelik açısından bildirilmiş çok az sayıda rapor mevcuttur. Biz bu yazıda bipolar bozukluk klinik bulguları ile başvuran ve takibinde nöbet gözlenen, tanı koydurucu deri ve nöroradyolojik bulgulara sahip ileri yaştaki bir hastayı tartışmak istedik. Şikayetleri, artmış benlik duygusu, uyuyamama ve paranoyalardı. Psikiyatrist tarafından değerlendirilen hastaya olanzapin ve lityum başlanmış, tedavisinin birinci haftasından itibaren yakınmalarında azalma gözlenmiş. Lityum tedavisini kesen hastada yine benzer yakınmalarla tekrar psikiyatri polikliniğine götürülmüş. Tedavisi sırasında hastada jeneralize tonik klonik nöbet gözlenmesi nedeni ile hastaya organik kökenli bipolar bozukluk ön tanısı ile kranial magnetik rezonans incelemesi (MRI) yapılmış. Nöroloji ile yapılan konsültasyon sonrasında lezyon atipik iskemik gliotik odak olarak değerlendirilmiş. Hasta 2 den fazla karakteristik klinik bulgu ve ek olarak radyolojik bulguları nedeni ile TSK olarak kabul edildi. Bu hastanın birkaç özel yönü olduğunu düşünüyoruz, bunlardan birincisi hastanın nörolojik yakınmalarının gelişmesinden çok önce başlayan psikiyatrik yakınmaları ve ikinci olarak semptomların çok ileri yaşlarda başlaması ve klasik triadda yer alan nöbetlerin çok geç kliniğe eklenmesidir.

Anahtar kelimeler: *Tuberoskleroz kompleksi, Bipolar bozukluk*

Abstract

Tuberous sclerosis complex (TSC) has many psychiatric comorbities like anxiety disorders, depression, autism spectrum disorder. However, bipolar disorder as a comorbidity to TSC has been rarely reported. In this article, we want to present a case who was initially diagnosed with bipolar disorder and then had a diagnosis of TSC. Her complaints were inflated sense of self, inability to sleep and paranoias. She was prescribed to olanzapin and lithium by a psychiatrist and she relieved from her symptoms after a while. Because she gave up her lithium treatment, her symptoms appeared again. During her treatment, she had a generalized tonic-clonic seizure. Cranial MRI was taken with a prior diagnosis of organic bipolar disorder. After consultation with neurology, the lesion was evaluated as atypical ischemic gliotic focus. The patient was diagnosed with TSC because she had more than two characteristic clinical signs of TSC together with radiological findings. This case presents two important points. First, psychiatric complaints occurred long before the development of neurological symptoms. Secondly, classical triads including seizures and the age of onset were too late compared to other patients with TSC.

Keywords: *Tuberous sclerosis complex, Bipolar disorder*

Introduction

Tuberous sclerosis complex (TSC) is a multisystem disease mainly characterized by triad of mental retardation, adenoma sebaceum and epilepsy [1]. TSC often comorbids to significant psychiatric disorders such as autism disorders spectrum, hyperkinetic disorders, depression and anxiety disorders. However, it has been rarely reported

Sorumlu Yazar: Tuba Özcan, Ordu Üniversitesi Tıp Fakültesi
Ordu Üniversitesi, Tıp Fakültesi, Nöroloji AD Cumhuriyet Yerleşkesi
dr_aydemir@yahoo.com

e-ISSN:2147-2181

that it comorbids to bipolar disorder [2-4].

TSC is a hamartomatosis which predominantly affects central nervous system, skin, retina, the kidneys and heart. Lungs, gastrointestinal tract, spleen, lymph nodes, gonads, endocrine glands, and bones may be affected to a lesser extent. In this autosomal dominant disorder, family history may not be taken in the three quarter of the cases [1-5].

Psychiatric disorders are common and varied in TSC. Clinical relationships between psychiatric disorders and the age of onset for TSC have been reported. In a retrospective evaluation, the most common comorbidity is anxiety disorders among adults, whereas autism spectrum disorders and attention-deficit hyperactivity disorder are the most common comorbidities among children. Mood disorders, especially bipolar disorder with manic episodes have been very rarely reported [3-5].

The most common neurological finding in TSC is seizure. The seizures have been reported to be the initial symptom in 75% of the patients with TSC [6-7]. The aim of the present article is to discuss an elderly case who had bipolar disorder diagnosis. Seizures were observed throughout her follow-up and she had diagnostically significant skin signs and neuroradiological findings.

Case Report

It was learned that 52-year old female patient with a bipolar disorder diagnosis had been followed for 5 years. Her husband presented her to the psychiatry polyclinic 5 years ago without any prior complaints. Her complaints were increased self esteem and inability to sleep. In addition to those, she had paranoias such as ; the local governors abused her, they made grandiose plans in order to maltreat her, soldiers and police wanted to keep her at home, and formed special forces to do so. Then she was started on olanzapin as far as learned from her history. It was observed that her complaints decreased as from the first week. She went back to her normal life by the third week. Lithium 600 mg/day was prescribed to her for her bipolar disorder diagnosis. The patient who gave up her lithium treatment after two years of her first symptoms began to have similar complaints. She was brought to the hospital and haloperidol (20 mg/day) was prescribed to the patient. Because he had a generalized tonic-clonic seizure at the third day of the treatment, cranial MRI was taken with a prior diagnosis of rule out organic bipolar disorder. T2 weighted hyperintense lesion was found in the right frontal region in cranial MRI. After consultation with neurology, the lesion was evaluated as atypical ischemic gliotic focus. It was expressed that the patient had a seizure because haloperidol reduced the seizure threshold. Haloperidol was given up and olanzapin was started. The patient's complaints expired after a while. She was started on lithium 600 mg/day and followed-up.

The patient under the lithium treatment was presented to us with the complaints of impairment in walking and having seizures. Before the application, she experienced a generalized tonic-clonic seizure. During the neurological examination we found spastic paraparesis. During the physical examination, we found sebaseum adenoma and subungual fibroma on the second finger of the left foot. There were also groups of yellow-colored maculopapular eruptions in abdominal and lumbosacral region, and confetti-style macules (Shagren patch) with the dimensions of 0.5x 1 cm in legs. In the electroencephalography of the patient, there were electrophysiological findings compatible with the organizational abnormalities which were generalized in both frontotemporal region and distinct in the left side, and also there were neuronal hyperexcitability. Her intelligence level was indicated as normal in Standard IQ tests. There were no individual with a story of any other psychiatric disorders and TSC in the patient's family. Thorax and abdominal CT was unremarkable. There was no pathological sign except left ventriculer hypertrophy in transthoracic echocardiography. Bone survey was normal. In cranial MRI there was a calsifying subependymal nodule in T2 weighted sequence near lateral ventricule. There were also several lesions which were hyperintense in T2 weighted and hypointense in T1 weighted sequences which the greatest was located on the right frontoparietal region and had dimensions of 1x2 cm compatible with cortical tubers (Figure

e-ISSN:2147-2181

1-2).

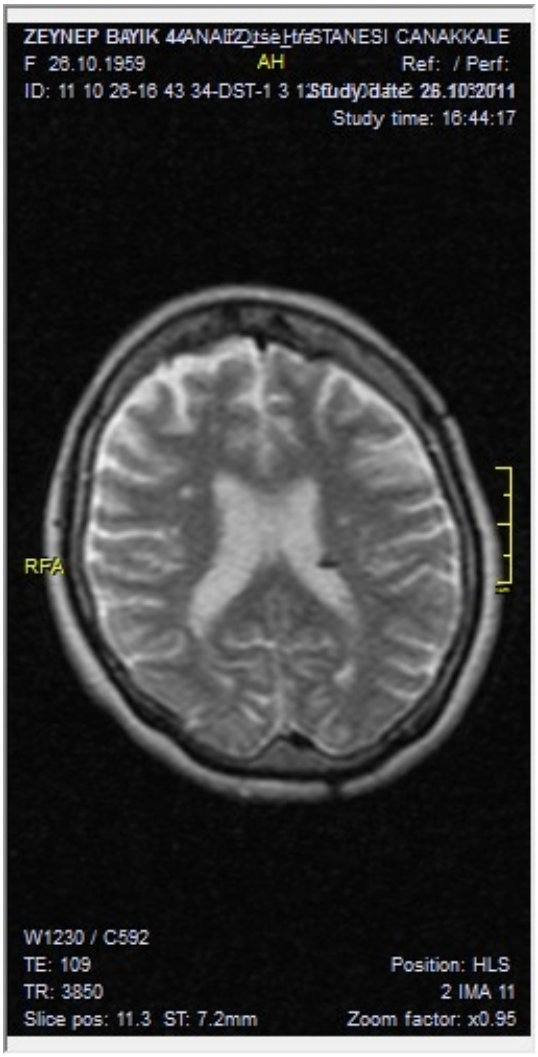

Figure 1
Calsifying subependymal nodule and lesions compatible with the cortical tuber

e-ISSN:2147-2181

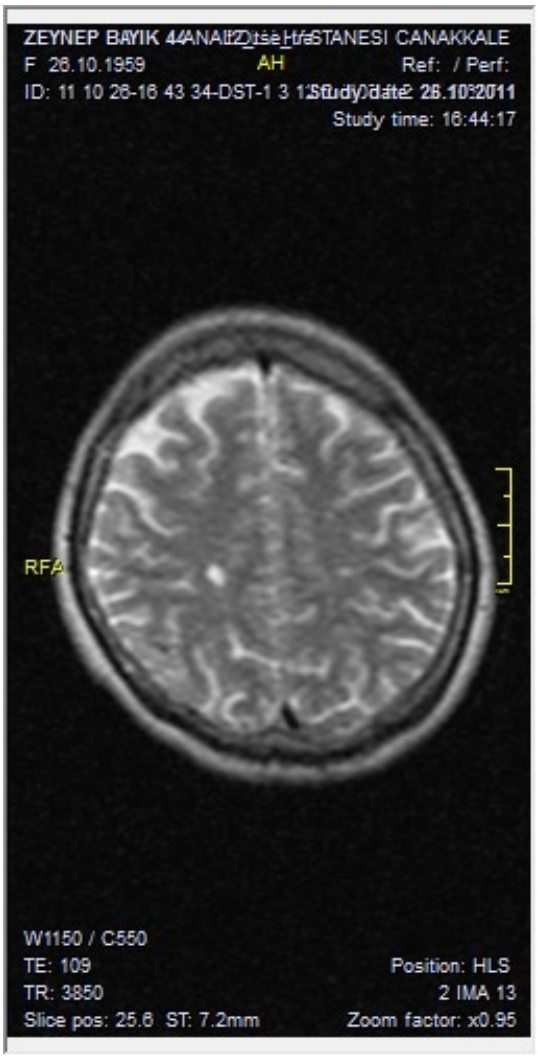

Figure 2
Calsifying subependymal nodule and lesions compatible with the cortical tuber

The patient was accepted as TSC because she had more than two characteristic clinical signs of TSC and additional radiological findings.

Discussion

We believe that this patient has several special aspects. Firstly, psychiatric complaints and symptoms, especially those developed as manic episodes based on bipolar disorder, started long before the development of patient's neurological symptoms and complaints. Secondly, late onset of the disease and late involvement of seizures which are in the classical triad of TSC were characteristic to this patient.

To the best of our knowledge, there was a report citing the co-occurrence of TSC and bipolar disorder with classic manic episodes. According to the report of Haq et al., the patient was as well diagnosed with classical bipolar disorder [4] like the patient of the present report. However, the patient had epileptic attacks before the manic episodes unlike our case. In our case, the first seizure of the patient was seen long after the first manic episode. Interestingly, compared to other TSC cases the age of onset was too late for our case. There was anyone with TSC in the patient's family and the progression of the disorder was too slow to make definite diagnosis. It is likely that

e-ISSN:2147-2181

the lack of mental deterioration caused the late onset seizures. There have been a lot of reports revealed the relationship between the age of onset of seizures and mental deterioration [5,6,8]. In our case, we were able to compare the images with the images captured before the diagnosis. In the pre-diagnosis images, there was a lesion which we interpreted as cortical tuber. In the latter images, there were increased number of cortical tuber and parenchymal lesions. These findings support the relationship between cortical tubers and seizures [2-8].

Right hemisphere lesions especially seem to be associated with bipolar disorder [9,10]. In our case, subcortical or cortical lesion load was also more in the right hemisphere than in the other. Subcortical lesions seem to be associated with manic-depressive episodes, whereas cortical lesions seem to be more associated with pure manic episodes [9,10]. When we reevaluated the MRI screening after the seizure in the second pure manic episode, we interpreted the lesion as cortical tuber. There have been studies revealing the relationship between TSC and psychiatric comorbidities with a genetic basis. It was reported that the different locus placement of terminal 2Mb of the short arm of chromosome 16 seemed to be related to psychiatric comorbidities [11], but it is claimed for broad psychiatric comorbidities.

As a result, in order to clarify the relationship between bipolar disorder and TSC, more case reports are needed. Even though it is very likely that there is a causal relationship between these two bipolar disorder may be an incidental comorbidity. We want to present this special case who had pure manic episodes and late onset TSC to reveal the relationship between bipolar disorder and TSC. We also want to indicate that bipolar disorder may be an indicator of TSC, rarely.

References

1. Hanno R, Beck R. Tuberous sclerosis. Neurol Clin. 1987;5:351-9.
2. Gillberg IC, Gillberg C, Ahlsen G. Autistic behavior and attention deficits in tuberous sclerosis: a population-based study. Dev Med Child Neurol. 1994;36:50-6.
3. Muzykewicz DA, et al. Psychiatric comorbid conditions in a clinic population of 241 patients with tuberous sclerosis complex. Epilepsy Behav. 2007;11:506-13.
4. Haq MZU, et al. Bipolar disorder and tuberous sclerosis complex: Is it a mere coincidence? CNS Spectr. 2009; 14 (11) :643-7.
5. Kingsley DPE, et al. Tuberous sclerosis: A clinicoradiologic evaluation of 110 cases with particular reference to atypical presentation. Neuroradiology. 1986; 28: 38-46.
6. Gomez MR. Neurocutaneous disease. In: Bradley WG, Daroff RB, Fenichel GM, Marsden CD (Eds.) Neurology in Clinical Practice. 2nd Edition, Volume II, Boston;Butterworth Heinemann: 1996;1566-71.
7. Nagib MG, et al. Tuberous slerosis: A review for the neurosurgeon. Neurosurgery. 1984;14:93-8.
8. Pampiglione G, Moynahan EJ. The Tuberous sclerosis syndrome: Clinical and EEG Studies in 100 Children. J Neurol Neurosurg Psychiatry. 1976;666-73.
9. Starkstein SE, Boston JD, Robinson RG. Mechanisms of mania after brain injury: 12 case reports and review of literature. J Nerv Ment Dis. 1988;176:87-100.
10. Starkstein SE, et al. Manic-Depressive and pure manic states after brain lesions. Biol Psychiatry. 1991;29:149-58.
11. Daniels RJ, et al. Sequence, structure and pathology of the fully annotated terminal 2Mb of the short arm of human chromosome 16. Hum Mol Genet. 2001;10:339-52.