CASE REPORT OF A SICKLE CELL DISEASE PATIENT WITH PRIAPISM TRIGGERED BY ANTI-CONVULSANT THERAPY IN JUTH

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ABSTRACT

Background: Priapism is defined as an abnormal persistent erection of the penis. It is usually painful and it is unrelated to sexual stimulation or unrelieved by ejaculation. Priapism results from a variety of possible etiological factors, including a number of pharmacological agents like antiepileptic medications.

Case summary: A 30-year-old man, a known sickle cell disease patient with background post cerebrovascular accident (CVA) seizure disorder, developed multiple episodes of priapism after commencement of a particular antiepileptic medication. The onset of priapism was noticed to coincide with the commencement of levetiracetam, an antiepileptic drug, and persisted while patient was on the medication.

He had several conservative and surgical interventions for the twelve episodes of priapism he had over the two-year period following commencement of levetiracetam. These included corporal aspiration, Winter's, T, Ebbehoj distal shunts with the last intervention being Al-Ghorab shunt. The withdrawal of the anticonvulsant, levetiracetam, however coincided with the cessation of priapism. He was subsequently commenced on Tab Epilin Chromo 200 mg bd, Meditriol 0.25 mgdly, and Neurovite forte 1 dly. He also became seizure free subsequently.

Conclusion: A possible causative factor of priapism in this patient, namely levetiracetam, could have been masked by the background hemoglobinopathy but for a high index of suspicion. The withdrawal of this anticonvulsant coincided with the resolution of priapism in this patient.

INTRODUCTION

Priapism describes a persistent penile erection arising from dysfunction of the mechanisms regulating penile tumescence, rigidity and flaccidity. It is defined as a full or partial erection that continues more than 4 hours beyond sexual satisfaction and orgasm or is unrelated to sexual stimulation.[1]

Possible etiological factors responsible for priapism include thromboembolic (sickle cell disease, leukemia, thalassemia, thrombocytopenia), neurogenic (spinal cord injuries, disc prolapse, cauda equina compression), medications (anticonvulsants, anticoagulants, antidepressants, antihypertensives, antipsychotics), malignancies, traumatic, and iatrogenic causes.[2]

Among anticonvulsants, the following medications have the potential for causing priapism: valpromide, brivaracetam, valproic acid, topiramate, oxcarbazepine,

clonazepam, carbamazepine and levetiracetam. Of these, valpromide has the largest association.[3] Bansal et al[4] reported a case of priapism associated with valproic acid. Perkoz et al[5] also reported a similar case in association with levetiracetam.

Rarely, there could be more than one possible risk factor in a patient with priapism. It is therefore necessary that all clinical armamentarium and acumen are brought to bear to ensure accurate identification of the offending risk factor.

Although, only a possible causality was established between levetiracetam and priapism, it is important that awareness is created by this report to help physicians in prescribing this drug safely in the future.

CASE SUMMARY

Mr. A.I is a 30-year-old who was diagnosed as a sickle cell disease patient in infancy and was regular on hematology outpatient clinic visits. His initial presentation to Urology Division of the above facility on account of priapism was three years ago with the first of many episodes of stuttering priapism. He went on to have eleven other episodes all managed accordingly. He has been free of the condition over four months from the time of review.

Prior to the initial presentation, he had multiple (five) episodes of right hemispheric ischemic cerebrovascular accident (CVA) spanning a period of twenty-one years with a sequela of psychomotor retardation. The first ever episode occurred when he was five years of age. He presented with the repeat episode to Jos University Teaching Hospital (JUTH) which was characterised by altered sensorium, slurred speech, impaired motor function and a ten minutes history of loss of consciousness. He is not a known hypertensive.

He subsequently developed multiple episodes (four) of seizure disorder. Seizures were generalized and tonic-clonic in nature with no preceding aura, no post-ictal sleep nor loss of sphincteric function. Each seizure episode lasted about 3-5 minutes. An electro-encephalogram done revealed multiple diffuse epileptiform activities. He was initially placed on Tab Carbamazepine 200 mg twice daily which was changed to Tab levetiracetam 500 mg daily when seizures became recurrent.

His first episode of low-flow priapism coincided with the commencement of levetiracetam in treatment of the seizure disorder. This occurred 27 years after being diagnosed as a sickle cell disease patient and seven weeks after the first episode of seizure which was initially treated with Carbamazepine. The episode resolved on non-operative treatment. Patient was discharged on oral analgesics, non-iron blood building supplements and advice on liberal oral fluid intake. He went on to have multiple (twelve) episodes of priapism spanning a period of two years.

INTERVENTION

The multiple episodes of priapism had warranted several conservative and surgical interventions including corporal aspiration, Winter's, T, Ebbehoj distal shunts, the last intervention being Al-Ghorab shunt with use of Hegar's dilator over the two-year period.

The recurrent nature of seizures which occurred intercurrently with episodes of priapism warranted the review of his anticonvulsant regimen. Oral levetiracetam was discontinued and patient was placed on Tab Epilin Chromo 200 mg bd, Meditriol 0.25 mg dly, and Neurovite forte 1 dly.

Episodes of priapism tailed off following withdrawal of Tab levetiracetam. Patient had no repeat episode of priapism after levetiracetam was discontinued and he also

became seizure free following review of anticonvulsant therapy. Better seizure control with Sodium valproate might have also had a role in resolution of priapism as the initial poor seizure control could have contributed to the prior recurrence of the priapism.

DISCUSSION

Priapism, a common urological emergency, usually arises from a variety of possible causes. Some etiological factors like hemoglobinopathies are more frequently implicated than others. It is a known fact that sickle cell disease is the cause of priapism in 2% to 29% of males with the disease.[6]

Among the less commonly reported causes of priapism are medications like anticonvulsants. Pekoz et al[5] reported a similar association identified in literature between levetiracetam and priapism. This was found in a 15-year-old male in Cukurova University, Adana, Turkey. Similar to our index case, the priapism resolved after withdrawal of the anti-convulsant.

The present case scored four on Naranjo adverse drug reaction probability scale[7], corresponding with a "possible causality". Since only a possible causality has been established, a definitive inference on this case is not warranted.

CONCLUSION

Even though the onset and cessation of episodes of priapism coincided with commencement and withdrawal of the anticonvulsant treatment for seizure disorder respectively, levetiracetam established only a possible causal association with priapism.

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