

Prolonged Penile Erection in an Adolescent with Angelman Syndrome under Aripiprazole: A Case Report

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ABSTRACT

Angelman syndrome (AS) is a genetic neurodevelopmental disorder characterized by mental retardation, ataxia, and happy social behavior. Aripiprazole is an atypical antipsychotic drug used for treating irritability in patients with autism spectrum disorder in > 6-year-old children and adolescents. Notably, prolonged penile erection is an unknown complication of Aripiprazole. Therefore, the present case report aimed to present a unique prolonged penile erection in an adolescent with Angelman syndrome under Aripiprazole. In this case report, priapism occurred several hours after consuming 5 mg of Aripiprazole. Since he was neither a cigarette smoker nor an alcohol abuser, had no medical issues, and did not report any history of penile or perineal trauma, the urologist suggested that the priapism he experienced might be linked to the use of Aripiprazole. Fortunately, he recovered after stopping the medication. Generally, we mentioned controversial results from previous case reports on the effect of Aripiprazole. It is unknown why some case reports noted priapism post Aripiprazole use, and some mentioned priapism palliation by switching to this drug from other antipsychotics. While it seems that the tendency to priapism may be an idiosyncratic reaction or the α 1-adrenergic receptors change or disorders in the predisposed patient, there is a need for further comprehensive studies on this issue.

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Introduction

Angelman syndrome (AS) is a genetic neurodevelopmental disorder that is characterized by mental retardation, ataxia, happy social behavior. This phenotype may overlap with autism (1). Spontaneous penile erection is a pathological condition, and its severe form presents as priapism. A urologic emergency manifests as a painful, spontaneous penile erection without any sexual stimulation. Although this condition is rare, it can lead to fibrosis of the cavernous bodies and, ultimately, erectile dysfunction (2). Priapism is classified into ischemic and non-ischemic types. The non-ischemic type occurs due to penile or perineal trauma. However, the ischemic type may happen as a result of diabetes, substance abuse, liver diseases, or hematologic disorders (i.e., sickle cell disease). Sometimes it has an iatrogenic cause (25-

40%) that usually arises regarding the administration of antidepressants, anxiolytics, antipsychotics, and anti-hypertension drugs. Among them, antipsychotic medications have the highest role (50%).

Aripiprazole is an atypical anti-psychotic drug that is used for treating irritability in patients with autism spectrum disorder in > 6-year-old children and adolescents (3). It has good oral absorption, and its effect peaks 3-5 hours after consumption. Additionally, the liver metabolizes it. The most common adverse drug reactions are somnolence, nausea, vomiting, esophageal dysmotility, vertigo, sialorrhea, tremor, and extrapyramidal complications (4). Notably, prolonged penile erection is an unknown complication of Aripiprazole, and rare case reports are available on this complication (3-8). This case report aimed to present a unique prolonged penile erection in an adolescent with AS under Aripiprazole.

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Case presentation

A 15-year-old boy, a known AS patient with the chief complaint of irritability and aggressiveness, was referred to the pediatric neurologist. The investigator prescribed Aripiprazole with a 5 mg starting dose as the preferred drug for these symptoms. He was referred back to the emergency department due to painful penile erection for some hours after consumption. The patient had no history of medical disease, as well as perineal or penile trauma, and did not take any medication, and was not a cigarette smoker or alcohol abuser. The pediatric neurologist then stopped the drug use and recommended urologic consultation.

No other urological disorder was present in this consultation, and fluid therapy was suggested. He became well in a short period with the fluid and supportive therapies. Considering the reduction of symptoms and lack of recurrence after stopping the drug, priapism caused by Aripiprazole was the probable diagnosis. Therefore, the drug was changed to low-dose Citalopram as an alternative, and the patient did not notice any complications on follow-up.

Discussion

Priapism is one of the rare complications of typical or atypical antipsychotics (9), which occurs due to blockage of $\alpha 1$ -adrenergic receptors in the cavernous body. Among older typical antipsychotics, Chlorpromazine and Thioridazine are more prone to block the receptor, as mentioned earlier. While the newer atypical antipsychotics such as Clozapine, quetiapine, and risperidone have the highest tendency (10), Aripiprazole has the lowest (5).

In this case report, priapism occurred for some hours after consuming 5 mg Aripiprazole. As he was not a cigarette smoker or alcohol abuser, he did not have any medical problem, he did not mention the history of penile or perineal trauma, and he recovered after drug cessation, the urologist indicated the possibility of priapism post-Aripiprazole. Notably, there are a limited previous reports on the occurrence of priapism by this drug as well (3-8). However, to the best of our knowledge, there is only one case report in children and adolescents. Negin et al. reported a 16-year-old adolescent with bipolar disorder. The patient had priapism two days after adding Aripiprazole to Oxcarbazepine and Lithium (6). Two significant differences existed between them in this study. Although they noted priapism post-polytherapy, our case underwent monotherapy. Furthermore, the studied patient had a genetic disorder and a psychiatric disease. Besides, some reports are available on adults with psychiatric disease. Mago et al. reported a 47-year-old man with schizophrenia who encountered

recurrent priapism after consuming Aripiprazole (5). There are also two case reports by Hsu et al. (3) and Aguilar et al. (4) on the relationship between the dosage of Aripiprazole and priapism. Hsu et al. demonstrated a 24-year-old man with psychotic disorder (8), and Aguilar et al. mentioned a 23-year-old man with schizophrenia (4). Two other reports were conducted by Togul et al. in a 30-year-old man (7) and Trivedi et al. in a 25-year-old men with schizophrenia (8). In contrast to reports mentioned above, Debby et al. mentioned the palliative effect of Aripiprazole on priapism in patients with schizophrenia who consumed other antipsychotics, and they concluded that in the case of priapism due to other antipsychotics, the clinicians should switch the treatment to Aripiprazole (2).

This case report had some unique points. Contrary to some previous reports, our report was not dose-dependent. This case occurred in the pediatric field, meaning typical risk factors like cigarette smoking, alcohol abuse, and age-related diseases were not present in this age group. Unlike others, this patient had a genetic disorder, but no psychiatric conditions such as schizophrenia or bipolar disorder were noted.

In Conclusion

Generally, we mentioned controversial results from previous case reports on the effect of Aripiprazole. It is unknown why some case reports noted priapism post Aripiprazole use, and some mentioned priapism palliation by switching to this drug from other antipsychotics. While it seems that the tendency to priapism may be an idiosyncratic reaction or the $\alpha 1$ -adrenergic receptors change or disorders in the predisposed patient, there is a need for further comprehensive studies on this issue.

Acknowledgment

We appreciate our colleagues for their cooperation. The parents approved the written informed consent letter. This study was ethically approved by the Vice-Chancellor of Research at Guilan University of Medical Sciences (Code: IR.GUMS.REC.1402.109).

Authors' Contribution

Afagh Hassanzadeh Rad: Made significant contributions to the conception and design of the study. She was involved in acquiring, analyzing, and interpreting the data. Additionally, she drafted the manuscript and gave her final approval to publish the version. She agrees to be accountable for all aspects of the work, ensuring accuracy and integrity.

Vahid Aminzadeh: He played a crucial role in the study's conception and design. He participated in the

data acquisition, analysis, and interpretation, contributed to drafting the manuscript, and approved the final version for publication. He takes responsibility for all aspects of the work, ensuring its accuracy and reliability.

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Conflict of Interest

The authors declared no conflict of interest.