Methylphenidate-induced priapism in a prepubertal boy

Sir,

Methylphenidate is a stimulant medication approved for pharmacological management of attention-deficit hyperactivity disorder (ADHD) in children above 6 years of age. Common side effects of methylphenidate use include insomnia, decreased appetite, weight loss, irritability, and anxiety. Tachycardia and hypertension have also been reported which may be dose related.

One of the uncommon side effects of methylphenidate use includes priapism, which is understood as a prolonged full or partial penile erection which is unrelated to sexual stimulus. Following the publication of around 15 case reports of priapism in children (mean age - 12.5 years) associated with the use of stimulants, the US Food and Drug Administration issued a warning in 2013, suggesting an association between priapism and stimulants (methylphenidate and atomoxetine).

Most of the cases of priapism seen with the use of methylphenidate have been observed in children aged 8 years or more and adolescents. In this report, we present a case of a 4-year-old male who presented with the symptoms of priapism and review the literature on the association of methylphenidate and priapism.

A 4-year-old boy was brought with the chief complaints of rubbing his penis repeatedly after started using methylphenidate. Detailed evaluation of his history revealed that he was born out of a wanted, planned pregnancy of nonconsanguineous marriage by in vitro fertilization. He was delivered by an elective cesarean section, without any antenatal or perinatal complication. He cried immediately after birth, his birth weight was ~ 2.2 kg, and breastfeeding was instituted immediately after birth. He developed transient physiological jaundice which resolved in 2–3 days without any phototherapy. He had delayed development of milestones in all domains of fine motor, gross motor, speech, language, and communication. He started walking by the age of 2½ years. By 3 years of age, when he was sent to play school, he was noticed to be restless, would not sit still even for 5 min, would be always on the go, keep fiddling with objects in his reach, playing with them for few minutes, and then throwing them away. At the play school too, he would not sit on his seat, instead kept on roaming in the entire class, disturb other children, despite repeated instructions of teachers to not to do so. If made to sit forcibly, he would keep on moving his limbs and appear restless. He would not pay attention to what was being taught in class, instead keep on looking outside the windows and doors. When teachers would try to engage him in any activity such as drawing, he would finish it fast and then interrupt other children in their work. He had difficulty in waiting for his turn while playing; often insist to let him play first. He would keep talking something or the other or singing a rhyme despite being told not to do so. He often interrupted others during conversations. In addition, he often lost his belongings at school. As a result, he was not able to learn and lagged behind his peers. Parents thought that things would improve as he grew but all these behaviors kept on increasing.

Due to his behavioral problems, there were frequent complaints from the school and this led to a psychiatric consultation. He was evaluated by a psychiatrist, was diagnosed with ADHD, and was started on tablet methylphenidate (immediate release) 5 mg/day in the morning. Over the period of the next 2–3 months, there was about 50% reduction in his hyperactivity. However, after about 3 months of continuing methylphenidate, the family members observed him to be rubbing his penis repeatedly, many times a day, each time for 5–10 min, especially few minutes to half an hour after being administered the morning dose of methylphenidate. When family members would ask him to stop, he would not stop and it would appear that he was unable to control it. His penis would appear to be erect, which would subside after 10–15 min, without any ejaculation. There was no associated history of any urinary symptom, sexual abuse, and exposure to any kind of sexually arousing materials prior to development of these symptoms. In view of the temporal correlation between the administration of methylphenidate and sexual symptoms, the family members tried to give methylphenidate during the afternoon (2 pm), with which symptoms would not be seen in the morning hours, but start within 2 h of administration of the dose and then would keep on occurring 3–4 times in the day. However, administration of methylphenidate during the afternoon time was associated with an increase in irritability, reduction in appetite, and reduction in sleep. In between, the family members experimented with stoppage of medication for few days, which would be associated with a lack of sexual symptoms but increase in the ADHD symptoms. Hence, they would reinstitute the medication.

On examination, his height and weight were appropriate for age. Head circumference (occipito-frontal circumference) was 50 cm. Local examination of genitalia revealed a flaccid penis and there was no erythema around the genitalia. During the interview, his behavior was consistent with the diagnosis of ADHD.
Based on the history and examination, a diagnosis of ADHD with the possibility of methylphenidate-induced priapism was kept. Methylphenidate was discontinued in view of the child's age (<6 years) and possibility of association of priapism with methylphenidate. He was managed with nonpharmacological measures in the form of structured routine and timetable, positive reinforcement, contingency management along with parental counseling, and psychoeducation. He showed an improvement in ADHD symptoms, his behavior improved in the class and at home, and his performance in studies also improved. There were no episodes of rubbing of genitalia and erection after stopping methylphenidate.

To the best of our knowledge, this is the first report of recurrent erections in a 4-year-old child associated with the use of methylphenidate. In most of the previously published case reports, priapism has been noted in adolescents or prepubertal boys. Existing literature suggests that priapism with methylphenidate can occur with both immediate release and long-acting formulations, may be related to the dose of the medication and can also occur during the period of drug withdrawal.[3] In many of the patients who developed priapism with methylphenidate, the dose used was much higher (10–54 mg/day) than that in the index case.[4][5] In the reported cases, management of priapism involved discontinuation of the offending agent, without any recurrence in the symptoms.[4][5] In the index case too, priapism did not recur after discontinuation of methylphenidate.

The mechanism of priapism associated with methylphenidate is thought to be mediated through the dopaminergic system.[8] From the existing literature and the reported case, it can be concluded that priapism can occur as a side effect of methylphenidate. Possibly, this side effect is usually not reported because of embarrassment in discussing such issue and lack of knowledge. Therefore, it becomes mandatory on the part of treating clinicians to inquire about this uncommon side effect. Further, whenever methylphenidate is prescribed to a male patient, the patient and caregivers must be informed about the signs and symptoms of priapism and what is to be done in the eventuality of the development of such symptoms.

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Conflicts of interest
There are no conflicts of interest.

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