have been Cushing's syndrome (due to the catabolic proteolytic effect of glucocorticoids upon the intestinal smooth muscle), together with chronic hypopotassemia, no other similar cases having been found in the literature. The partial response after bilateral adrenalectomy supports the hypothesis of chronic damage established upon the colon wall.

In sum, we have reported a case of hypercortisolism associated with a megacolon, the cause of which may be related to the structural damage induced by cortisol overproduction and chronic hypopotassemia.

## Conflicts of interest

The authors declare that they have no conflicts of interest.

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# Priapism associated with cabergoline in a young adult☆

## Priapismo asociado al uso de cabergolina en un adulto joven

Prolactinomas are the most prevalent pituitary adenomas. While these lesions are more common in women (gender proportion 10:1), in males they tend to manifest in the form of macroprolactinomas. Although such lesions may manifest through a mass effect as headaches and vision disturbances, central hypogonadism is the main manifesting syndrome regardless of tumour size. Dopamine agonists such as cabergoline constitute an effective treatment for prolactinomas, achieving a normalization of prolactin levels and a decrease in tumour size. Their side effects include cardiac valve alterations and, less commonly, compulsive behaviour and hypersexuality.[1] Only one case of priapism as a side effect of cabergoline has been reported in the literature to date.[2] We report another case in a young adult with macroprolactinoma.

An 18-year-old male with a history of type 1 diabetes mellitus since 10 months of age presented with a weight gain of 10 kg over the past year (weight 79.5 kg, height 1.70 cm), incomplete pubertal development (Tanner stage III–IV, testicular volume 12 cc) and a bone age of 16.5 years. The laboratory tests revealed hypogonadotropic hypogonadism with total testosterone levels of 2.41 ng/mL (2.6–10 ng/mL), LH 1.68 IU/L, and FSH 3.33 IU/L. Hyperprolactinemia (192 μg/L) was also observed, with all other pituitary hormone levels and IGF-1 within normal ranges (228.6 ng/mL). Magnetic resonance imaging showed the presence of a macroadenoma measuring 12 × 9 mm in size, with no suprasellar extension, causing contralateral displacement of the pituitary stalk, in the absence of headache and vision disturbances in confrontation visual field examination. Cabergoline 0.25 mg twice weekly was started as treatment.

After six months of treatment, the patient showed a marked decrease in prolactinemia (0.59 μg/L) associated with clinical (Tanner stage IV–V, testicular volume 16 cc) and laboratory test pubertal development (total testosterone 8.89 ng/mL [2.6–10 ng/mL], LH 3.43 IU/L, and FSH 5.14 IU/L), as well as a weight loss of 8.6 kg and a 1-cm increase in height. Despite the good clinical course, over the following months he experienced weekly episodes of painful, very prolonged erections, always in the absence of sexual stimulation, and had to report to the emergency

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room up to 6 times for lack of spontaneous detumescence after several hours. Aspirated corpus cavernosum blood gas testing revealed acidosis, hypercapnia and hypoxia suggestive of ischemic priapism. Reversal of the priapism episodes required intracavernous sympathomimetic agents (epinephrine or ephedrine), and blood aspiration in some instances. The patient denied any use of recreational drugs or other medications apart from cabergoline and his regular treatment in the form of subcutaneous multiple dose insulin.

An outpatient study by the reference urologist, with Doppler ultrasound of the cavernous arteries, yielded no relevant findings, the case being oriented as priapism probably secondary to cabergoline. Given this situation, and after completing one year of treatment, with low prolactin levels and tumour reduction (7 mm), the decision was made to reduce the cabergoline dose to 0.25 mg/week. Following the dose reduction, the patient experienced weekly episodes of painful erections lasting less than an hour and with spontaneous detumescence over the following months, without having to again visit the emergency room. In the presence of low serum prolactin (0.66 ng/mL), the cabergoline dose was reduced to 0.125 mg/week, followed by no further episodes of priapism and the maintenance of low prolactin levels (6.96 ng/mL) and radiological stability (6 mm).

Ischemic or low-flow priapism is regarded as a urological emergency. If sustained over time, it may cause structural damage to the erectile tissue, with necrosis and fibrosis of the corpora cavernosa. For this reason, a rapid diagnosis is required, with suppression of the possible triggering factors. Priapism may occur as a side effect of different drug treatments, the most common being those used for erectile dysfunction. There have also been reports of cases associated with the use of antidepressants, antipsychotics and antihypertensive drugs. In the reported cases of drug-induced priapism, no associations have been found with the dosage or the duration of the treatments, priapism being able to manifest after the first dose or after a period of treatment. Our patient had been treated with cabergoline for 6 months.

In addition, due to the evident time coincidence and improvement following cabergoline dose reduction, the suspected cause-effect relationship is based on biological plausibility, since dopamine is a key neurotransmitter involved in erection and the central control of sexual function, acting upon the hypothalamic paraventricular area.[3] In fact, apomorphine, a dopaminergic $D_1/D_2$ receptor agonist, is useful for the management of erectile dysfunction.

Although we have only found one previously reported case of priapism secondary to cabergoline,[2] there are several documented cases involving short half-life and the daily transdermal administration dopamine agonists used in Parkinson's disease, such as rotigotine.[4] A case of clitoral priapism secondary to bromocriptine has also been reported in a female patient.[5]

In the present case, priapism could also be related to the increase in testosterone levels experienced by the patient after starting cabergoline. Although it has been described in the course of treatment with testosterone,[6] we have found no cases of priapism after the resolution of hypogonadism in patients with hyperprolactinemia.

Although priapism is a very uncommon adverse effect of cabergoline, we consider it relevant to report this case in order to enhance awareness of this association and thus facilitate earlier diagnosis and treatment.

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