Spinal Cord (2004) 42, 134–135

www.nature.com/sc

Letter to the Editor

Spinal Cord (2004) **42,** 134–135. doi:10.1038/sj.sc.3101547

Management of recurrent priapism in a cervical spinal cord injury patient with oral baclofen therapy

Priapism is a persistent penile erection that is unrelated to sexual stimulation. Subtypes of priapism include ischaemic, nonischaemic and stuttering. Stuttering or intermittent priapism is a recurrent form of ischaemic priapism in which penile erections occur repeatedly with intervening periods of detumescence. Although each episode of stuttering priapism may last only for a short duration with spontaneous penile detumescence, the condition challenges the physician to develop a management strategy for the prevention of future episodes of priapism. Treatment modalities for patients with stuttering priapism include systemic therapies, early self-injection of sympathomimetic agents (eg phenylephrine) into the corpus cavernosum and, as a last resort, surgical placement of a penile prosthesis. Among the systemic therapies, hormonal agents, baclofen, digoxin and terbutaline have been used to prevent stuttering priapism although the American Urological Association Guidelines for management of priapism do not recommend the use of terbutaline or digoxin for prevention of stuttering priapism. At present, there is little evidence to support the use of either terbutaline or digoxin in this clinical setting.[1] Hormonal therapy for stuttering priapism suppresses serum testosterone levels by feedback inhibition (diethylstilbestrol), blocking androgen receptors (antiandrogens), or by downregulation of the pituitary gland (gonadorelin analogues). We describe management of a patient with cervical spinal cord injury, who developed recurrent episodes of penile erection.

A 46-year-old male sustained C-4 complete tetraplegia in a mountain bike accident. CT scan showed burst fracture of C-4 vertebra, and fracture of arch of C-1. There was no evidence of trauma to pelvis or perineum. He underwent C-4 vertebrectomy, grafting and plating. He was sedated and ventilated through a cuff tracheostomy tube, when he was transferred to the spinal unit about a month after the accident. During his stay in the spinal injuries unit, he was weaned off the ventilator. Later, the tracheostomy tube was removed. He had a size 12 Fr. Foley catheter per urethra for drainage of urinary bladder. Medications included salbutamol nebuliser four times a day, omeprazole, carbamazepine, mianserin, senna syrup, bisacodyl rectal solution, and dalteparin sodium for prevention of deep-vein thrombosis.

In this patient, by the time of 12 weeks after the acute injury, movement of indwelling urethral catheter, for example, while turning from side to side or, changing the bed sheets triggered erection of penis. Initially, penile erection lasted only for short periods of 5–10 min. After 2–3 days, the penis stayed erect for increasingly longer periods up to 30 min. Although these episodes subsided spontaneously, erection of penis was embarrassing to female carers and to the patient's partner. Further, the indwelling urethral catheter acted as a bowstring and tended to erode the urethra when penis became erect. Therefore, we decided to treat recurrent priapism.

Full blood count was normal except for a slightly low haemoglobin value of 11.8 g/dl. Colour duplex ultrasonography of penis showed no anatomical abnormalities, such as a cavernous artery fistula or pseudoaneurysm. This patient was prescribed baclofen 10 mg, three times a day. After 24 h of therapy with baclofen, erection of penis occurred less frequently and each episode lasted for shorter duration. Three days later, nurses could turn him from side to side or change the bed sheets without triggering erection of penis. There was no side effect from baclofen such as sedation. This patient continues to take baclofen 10 mg three times daily for prevention of penile erection. It is likely that he may develop spasticity, when the dose of baclofen will require to be increased, perhaps to 20 mg four times a day.

Baclofen is an agonist of gamma aminobutyric acid receptor. Several studies in rats and men have suggested that baclofen inhibits penile erection and ejaculation.[2] Denys *et al*[3] studied nine men with spinal cord injury or multiple sclerosis, who were receiving intrathecal baclofen by an implantable pump; average follow-up was 44.4 months. Eight patients reported a decrease of erection rigidity and/or duration subsequent to intrathecal baclofen therapy. These authors concluded that intrathecal baclofen may compromise penile erection and ejaculation, but that this effect is reversible.

Very few patients have been treated with oral baclofen for treatment of recurrent priapism. Rourke *et al*[4] treated a 41-year-old male, who suffered from recurrent nocturnal priapism. The authors initiated treatment at a dose of 10 mg given at bedtime, with dose escalation in 10-mg increments. Complete alleviation of symptoms was achieved with a dose of 40 mg. Although baclofen therapy controlled the episodes of recurrent priapism, this patient's normal sexual function was preserved. The response to baclofen lasted during a follow-up period of 12 months. Priapism recurred when attempts were made to reduce the dose of baclofen or, when the patient did not take baclofen inadvertently. In another patient (age: 35 years) with megalopenis and recurrent episodes of idiopathic nocturnal priapism, Rourke *et al*[4] prescribed a 40 mg dose of baclofen at bedtime, which resulted

in complete cessation of unwanted erections with preservation of normal sexual function.

We preferred oral baclofen therapy to oral digoxin or terbutaline, as digoxin and terbutaline may produce serious cardiac side effects. We were reluctant to prescribe diethylstilbestrol in this patient with C-4 tetraplegia, as diethylstilbestrol might increase the risk of deep-vein thrombosis. Dahm *et al*[5] prescribed an antiandrogen, bicalutamide 50 mg once every other day to a 30-year-old male with stuttering priapism. This patient sustained back injury 3 years previously and developed chronic back pain, which was treated successfully by implantation of an epidural spinal cord stimulator. During a follow-up of 2 years, this patient noticed no changes in his libido or problems in achieving an erection while taking bicalutamide. The cost of bicalutamide 50 mg once every other day, if taken continuously for a year, will amount to sterling 834 pounds. The cost of baclofen 10 mg, taken three times a day for 1 year, is only sterling 30 pounds. Thus, treatment of stuttering priapism with oral baclofen is certainly cost-effective.

The ability to control stuttering priapism with oral baclofen leads to the interesting speculation that idiopathic priapism may be primarily a neurological disease.[2] It is possible that stuttering priapism may represent an early warning sign of a systemic neurological disease. Therefore, longitudinal follow-up of patients with stuttering priapism might be recommended to determine the natural history.

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