Case Report

The Use of Imipramine in a Child with Hinman's Syndrome: Case Report

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Background: *The Hinman's syndrome is a serious and unusual outcome of the dysfunctional elimination syndrome. The data about treatment of this syndrome is very limited.*
Objective: *To describe an unusual treatment of the Hinman's syndrome in a child by using imipramine.*
Material and Method: *A 7-year-old Thai boy presented with both diurnal and nocturnal enuresis and dribbling for one year. After many investigations were performed, the diagnosis of Hinman's syndrome was established. Finally, imipramine was used for the treatment.*
Results: *The patient had a good response to the treatment by imipramine. Before the treatment was started, he had both diurnal and nocturnal enuresis almost every day. After one month of treatment, he became dry 4 to 5 days/week in daytime and 5 to 7 days/week at night.*
Conclusion: *This report shows that imipramine may play a role in the treatment of Hinman's syndrome.*

Keywords: *Hinman's syndrome, Enuresis, Imipramine*

J Med Assoc Thai 2011; 94 (4): 515-7
Full text. e-Journal: *http://www.mat.or.th/journal*

The Hinman's syndrome is a serious and unusual outcome of the dysfunctional elimination syndrome. In 1973, Frank Hinman described the non-neurogenic neurogenic bladder in 14 boys approximately 8-years-old who had incontinence, infections and radiographic abnormalities of obstructive uropathy[1]. Since then, several articles have been published on this syndrome and its treatment.

The syndrome usually affects children in school age. They usually present with urination difficulty, enuresis, incontinence, urinary tract infection, or kidney failure[2]. Associated problems such as behavioral and psychosocial problems and bowel dysfunction, especially constipation are also seen in this syndrome[2]. In addition, videourodynamic findings may vary from mild detrusor overactivity and little postmicturition residue to severe bladder dysfunction associated with end stage renal failure[2]. The most typical finding is functional bladder outlet obstruction or detrusor sphincter dyssynergia[2]. The diagnosis is made by exclusion of neurological pathology. The etiology is actually unknown but factors including genetic predisposition, recurrent urinary tract infections, immaturity or too-early toilet training and sexual abuse are proposed[3].

Treatment modalities used in dysfunctional voiding include biofeedback, behavioral therapy, and medication[3-6]. Anticholinergic drugs and alpha-adrenergic blockers are often used in pediatric diurnal enuresis. Imipramine is a tricyclic antidepressant but also used in treating pediatric enuresis especially nocturnal enuresis.

Case Report

A 7-year-old Thai boy presented with both diurnal and nocturnal enuresis and dribbling for about one year before coming to Ramathibodi Hospital. Although he became dry during daytime at the age of three, he was never dry at night. He did not have constipation. During his course of illness, he had suffered from urinary tract infection three times.

His physical examination including neurological evaluation was normal. He did not have phimosis. Urine analysis and blood analysis for renal function were normal. Ultrasound of kidney-urinary bladder system showed mild hydronephrosis of both kidneys and diffusely irregular thickening of the bladder mucosa with increased vascularity. No

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demonstrable cause of obstruction was found. Voiding cystourethrogram showed trabeculation with multiple diverticula at the urinary bladder. Stricture at membranous urethra and meatus was considered. Magnetic resonance imaging of his spine was normal but bilateral mild hydronephrosis was observed. Urodynamic evaluation showed detrusor-sphincter dyssynergia.

He had no family history of enuresis. He was born by vacuum extraction because during the perinatal period, his mother had fever that gave her fatigue. His birth weight was 3,100 g and his developmental profiles were quite normal except his language development was reported delayed. He got his first meaningful word when he was about two years old. During school age, he was a cheerful boy with good performance at school and had no behavioral problem. However, his mother worried about his symptoms and she tried to control her son to go to the toilet.

The treatment with oxybutynin, anticholinergic agent, 5 mg by oral route two times a day (0.1 mg/kg/dose) was initiated. Along with medication, dilatation of urethra was performed. His urethra was dilated by sound number 2/5 to 3/6 without difficulty. However, he had no response to the treatment. Then doxazosin, alpha-blocker, 2 mg by oral route once a day (0.05 mg/kg/dose) was used instead of oxybutynin but he also had no response to this regimen. He always wore diapers.

After three years of treatment, the boy was referred to child psychiatrist. Imipramine 25 mg by oral route at night was tried. He had a good response to the treatment. Before the treatment was started, he had both diurnal and nocturnal enuresis almost every day. After one month of treatment, he became dry 4 to 5 days/week in daytime and 5 to 7 days/week at night. Then, dose increasing to 50 mg was tried but he could not tolerate the side effect, which is somnolence. Therefore, the dose was decreased back to 25 mg and behavioral modification was initiated. Positive reinforcement techniques such as token economy were used along with the medication.

Discussion

Until today, the certain etiology of Hinman's syndrome is unknown. Several methods are used to avoid renal deterioration including clean intermittent catheterization, bladder training and medications. There are some successful reports in the treatment of this syndrome with anticholinergic agents and alpha-adrenergic blockers but in case of imipramine, data is very limited[2,3]. However, imipramine has been used in treatment of nocturnal enuresis for a long time. The mechanism behind imipramine's antienuretic effect is debated. Apart from the drug's known weak anticholinergic effect, both effects on arousal, sphincter tone and urine production have been suggested[4].

In this case, anticholinergic agent and alpha-adrenergic blocker were tried before using imipramine but with no response. Therefore, a mechanism involving in the response may be other effects except anticholinergic effect. However, a natural course of the syndrome is unknown, so we cannot eliminate the effect of the syndrome per se. Moreover, the permanent effect of imipramine treatment is also unknown.

Due to the side effect of imipramine, dose titration is limited in the present case. In general, adverse effects of imipramine are uncommon at low dose and include gastrointestinal disturbances, sleep disturbances, anxiety, and dry mouth[4]. Most serious adverse effect is cardiotoxicity. Moreover, some authors mentioned about psychological stresses contribute the symptoms[3]. In the present case, the authors cannot identify the psychological stressor during symptom development. After the treatment, there was a mild interpersonal conflict between the mother and son. However, the symptom improvement occured before the authors manipulated this conflict by using positive reinforcement techniques.

Conclusion

The Hinman's syndrome is a serious and unusual outcome of the dysfunctional elimination syndrome. It can lead to renal insufficiency or chronic renal failure. This syndrome is diagnosed by exclusion of other neurological and anatomical disorders. This report shows that imipramine may play a role in the treatment of this syndrome.

Potential conflicts of interest

None.

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รายงานผู้ป่วย: การรักษาผู้ป่วยกลุ่มอาการฮินแมน ด้วยยาอิมิพรามีน

นิดา ลิ้มสุวรรณ

ผู้ป่วยเด็กชายอายุ 7 ปีมาพบแพทย์ด้วยอาการปัสสาวะราดทั้งในเวลากลางวันและกลางคืน นอกจากนั้นยังมีอาการปัสสาวะหยดออกมาโดยควบคุมไม่ได้ จากผลการตรวจทางภาพถ่ายรังสี การฉีดสีตรวจกระเพาะปัสสาวะและท่อไต รวมถึงการตรวจการทำงานของระบบขับถ่ายปัสสาวะ พบว่าผู้ป่วยมีลักษณะเข้าได้กับกลุ่มอาการฮินแมน หลังจากการรักษาด้วยวิธีอื่นไม่ได้ผล ยาอิมิพรามีนได้ถูกนำมาใช้รักษาผู้ป่วยรายนี้พบว่าผู้ป่วยมีการตอบสนองต่อการรักษาด้วยยาอิมิพรามีนเป็นอย่างดี