A Rare Cause of Urinary Retention in Women: Fowler’s Syndrome

Cihan Kaya*, Hediye Dagdeviren, Caglar Helvacioglu and Huseyin Cengiz
Department of Obstetrics and Gynaecology, Bakirkoy Dr Sadi Konuk Training and Research Hospital, Saglik Bilimleri University, Turkey

*Corresponding author: Cihan Kaya, Department of Obstetrics and Gynaecology, Bakirkoy Dr Sadi Konuk Training and Research Hospital, Saglik Bilimleri University, Istanbul, Turkey, Tel: 905536161385; E-mail: drchankaya@gmail.com

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Abstract

We present a rare cause of urinary retention. Fowler’s syndrome affects young women with painless urinary retention and as there is no known cure, the main concern is to ensure adequate bladder emptying. Our patient went through self-catheterisation for eight weeks and after this treatment her urethral output increased gradually and bladder residual volume was observed as <100–150 ml. Early investigation of chronic urinary retention and appropriate treatment can result in better improvement in their quality of life.

Keywords: Clean intermittent catheterisation; Painless urinary retention; Residual urine

Introduction

Chronic urinary retention (CUR) is a condition with non-painful bladder and chronic high post-void residual urine volume [1]. CUR may be due to decreased detrusor activity or bladder outlet obstruction. Fowler’s syndrome (FS) is a type of CUR which is seen in young women and it was described as a unable to void with absence of any described organic pathology [2]. Typically such women present with retained urinary volume over a litre, but had impaired sensation of bladder fullness. In most of those patients, no underlying neurologic, metabolic or other medical diseases can be identified which lead to inability to relaxation of external urethral sphincter [3].

Many of these women usually have a trigger such as urinary tract infection, general anaesthesia, childbirth or gynaecologic surgery [4]. Further, there are many numbers of associations between FS and gynaecological conditions with a hormonal basis such as polycystic ovarian syndrome (PCOS), endometriosis, ovarian cysts and pregnancy [5].

In this paper we described a patient with FS after transobturator tape procedure (TOT) and presence of PCOS.

Case Presentation

A 35 year old multiparous woman was admitted to our gynaecological clinic with a complaint of stress urinary incontinence (SUI). She had a history of ovarian cystectomy five years ago and on her gynaecologic ultrasound examination there were more 12 follicles in each ovary with increased ovarian volume. Specific evaluations of SUI such as the pad test for urine leakage, Q-tip test and pelvic examination for pelvic organ prolapse were performed preoperatively. Preoperative urodynamic studies were performed included uroflowmetry and measurement of postvoid residual (PVR), cystometrogram, pressure-flow study and measurement of urethral closure pressure and valsalva leak point pressure. The urodynamic study was performed with an MMS International model solar silver urodynamic system (MMS International, Dover, NH, USA). In urodynamic study, there was a leakage due to an increase in abdominal pressure. A TOT operation was performed under general anesthesia. The Monarc Subfascial Hammock System (American Medical Systems, Minnetonka, MN, USA) was used as vaginal tape. At the end of the operation, a 16-Fr Foley catheter was indwelled and was removed the day after. After removal of urethral catheter PVR measured about 1500 ml. Foley catheter was indwelled again. Cystourethroscopy was performed for evaluation of suspected operative urinary tract injury (e.g., ureteral injury, cystotomy, intravesical placement or erosion of mesh or suture). There were no pathology both in urethra and bladder. Urodynamic studies were performed again. There were post-operative urodynamic findings include reduced bladder sensation, increased bladder capacity and decreased detrusor activity. Clean intermittent catheterization (CIC) was recommended. After 8 week her urethral output increased gradually and bladder residual urine was measured as <100–150 ml.

Discussion

FS was first described by Professor Clare J in young women with painless urinary retention in 1985 [6]. The etiology of the syndrome is still remains unknown but it has been speculated that the disorder is due to a hormonally sensitive channelopathy lead to an inhibitory effect on detrusor contractions as well as the desire to void. As a consequence, the sphincter becomes overactive and hypertrophic and reacts excessively to direct stimulation.

The typical presentation is seen in woman aged between 20–30 years with an intermittent inability to void. This may happen spontaneously or may be after urinary tract infection, general anaesthesia, childbirth or a gynaecologic operative procedure [7,8]. Patients present with pelvic pain and fullness at lower abdomen. There are no defined histories of neurologic diseases and investigation of neurologic system is usually normal. It is important to exclude occult or undiagnosed neurological problems as an etiological cause. Our case was a 35 years old woman and there was no abnormal finding on her neurologic examination.

Over 80% of patients with FS had one or more gynaecological disorder. It is reported that patients with FS were thought to have a strong association with PCOS. Our patient had a history of ovarian cystectomy and also she had clinical and laboratory features of PCOS.
In the study by Fowler et al. FS was found to be associated with PCOS in 64% of patients but the cause of this association was unclear [9].

In those patients PVR volume is at least 1 litre, there is usually raised maximum urethral closure pressure on urethral pressure profilometry, increased sphincter volume on ultrasound or MRI assessment and a characteristic urethral sphincter electromyogram (EMG). The gold standard investigation is a urethral sphincter EMG [10]. There is an abnormal activity on EMG which causing outflow obstruction of the bladder and increased residual volume of urine. We offered performing EMG as a diagnostic test but, the patient did not want this diagnostic evaluation.

Conclusion

The aim of treatment is to try to ensure bladder emptying. In patients with low residual volume, no further intervention is necessary. But in patients with larger residual volumes, there should be a more definitive treatment, like intermittent self-catheterisation or placement of a suprapubic catheter. For women with more severe urinary retention and voiding dysfunction secondary to urethral sphincter over activity sacral nerve stimulation has shown to be the only effective therapy [10].

Early investigation of chronic urinary retention, correct diagnosis and appropriate treatment in this small group of women can result in better health and improvement in their quality of life.

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