Case Report

Refractory nocturnal enuresis associated with bulbar urethral stricture successfully treated by non-transecting urethroplasty

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Abbreviations & Acronyms
DVIU = direct vision internal urethrotomy
EPA = excision and primary anastomosis
Max Pves = maximal intravesical pressure
NTU = non-transecting urethroplasty
Q max = maximal flow rate shown by uroflowmetry
VCUG = voiding cystourethrogram

Introduction: Urethral obstruction has been reported to be involved in the pathogenesis of nocturnal enuresis, but such patients have been treated only endoscopically. We report a case in which nocturnal enuresis was successfully treated by an urethroplasty.

Case presentation: A 13-year-old boy was referred to our hospital for nocturnal enuresis, which was refractory to desmopressin acetate, anticholinergic drugs, and alarm therapy. Video urodynamic study findings showed bladder outlet obstruction associated with a short bulbar stricture. Two attempts to relieve the obstruction by direct vision internal urethrotomy improved enuresis, albeit transiently. A non-transecting urethroplasty was performed at the age of 15 years, which resulted in dramatic and durable improvements in enuresis, maximal flow rate in uroflowmetry, as well as other urodynamic parameters.

Conclusion: Urethroplasty, preferably by a non-transecting technique, can be a viable treatment option for intractable nocturnal enuresis associated with non-traumatic and short bulbar urethral stricture upon completion of puberty.

Key words: nocturnal enuresis, urethral stricture, urethroplasty.

Keynote message
Urethroplasty, preferably by a non-transecting technique, can be a viable treatment option for intractable nocturnal enuresis associated with non-traumatic and short bulbar urethral stricture upon completion of puberty.

Introduction
Standard initial treatment measures for monosymptomatic nocturnal enuresis in children are pharmacotherapy and alarm therapy, although a more specialized approach is recommended if there are daytime voiding symptoms or if the patient is refractory to such initial treatments. The involvement of bladder outlet obstructions such as urethral ring stricture and mini-posterior urethral valves has been reported in some cases, and are considered to possibly benefit from endoscopic incision. However, an urethral stricture in adults is considered to be an indication of open urethroplasty when endoscopic incision is ineffective or inappropriate.

Case presentation (Fig. 1)
A 13-year-old boy was referred to our hospital for refractory nocturnal enuresis. When he was 9 years old, he had been treated with desmopressin acetate, anticholinergics, and alarm therapy by a pediatrician specializing in enuresis, but all attempts had failed. The patient had no history of urethral trauma. Enuresis was present nearly every night, while there was no daytime incontinence. The dysfunctional voiding symptom score was 1/30 points, and the initial diagnosis was refractory monosymptomatic enuresis. At the time of the first visit, the genitalia was prepubertal.

Uroflowmetry showed a relatively flat flow pattern with a Q max of 13.0 mL/s for a voided volume of 118 mL, and post-void residual urine was 36 mL. A video urodynamic test indicated a normal storage phase with no uninhibited contractions, but bladder outlet obstruction
was demonstrated in the voiding phase, with an elevated Max Pves of 150 cmH2O, and cystourethrogram showed a 3-mm long stricture at the bulbar urethra (Figs 2a and 3a).

The patient was diagnosed with high-pressure urination associated with a non-traumatic and short bulbar urethral stricture and underwent an initial DVIU procedure at the age of 13 years and 11 months. Immediately thereafter, enuresis improved, and $Q_{\text{max}}$ was increased to 27 mL/s. However, 4 months later, both the enuresis symptoms and $Q_{\text{max}}$ worsened again in conjunction with stricture recurrence (Fig. 2b). A second DVIU was performed at the age of 15 years, which improved both enuresis and $Q_{\text{max}}$ similar to the first DVIU, while 4 months later, both had worsened. Enuresis persisted and remained refractory to medication. Puberty was completed, and it was unlikely that the stricture would improve by another DVIU or with further growth of the urethra at this age. Thus, an urethroplasty by NTU technique was performed: the bulbar urethra was incised longitudinally and horizontally sutured.

Following the procedure, enuresis was resolved without medication, and $Q_{\text{max}}$ did not worsen again. Max Pves remained at >100 mmHg after repeated internal urethrotomy, but at 4 months after urethroplasty, it decreased to 50 mmHg along with maintained urethral caliber shown by voiding urethrogram (Fig. 2c). The patient confirmed no erectile dysfunction during follow-up visits.

**Discussion**

In the present case, urethroplasty resulted in a dramatic resolution of nocturnal enuresis. Urethral obstruction has been reported to be involved in the pathogenesis of nocturnal enuresis, and such patients have been treated endoscopically. However, several of the older studies on this issue were conducted when conservative treatment with the behavioral therapy, or urotherapy approach was still uncommon.

![Fig. 1](image1) Clinical course of the present case.

![Fig. 2](image2) (a) VCUG findings and uroflow curve ($Q_{\text{max}}$ 13 mL/min) after initial visit. (b) VCUG findings and uroflow curve ($Q_{\text{max}}$ 17 mL/min) after first DVIU. (c) VCUG findings and uroflow curve ($Q_{\text{max}}$ 45 mL/min) 6 months after urethroplasty (NTU). The stricture was resolved after the urethroplasty procedure, in conjunction with improvement in enuresis.
More recently, one group demonstrated that transurethral ablation of the posterior urethral valve ameliorated diurnal incontinence and enuresis in boys aged 6–13 years old, using pressure-flow studies. However, patients in those studies were all treated endoscopically, while the present case is the first report in which nocturnal enuresis was successfully cured by urethroplasty.

This patient was refractory to standard treatments for several years, which may have been too long; thus, a video urodynamic test was performed immediately following his initial visit, and involvement of a non-traumatic and short bulbar urethral stricture was diagnosed. The improvement of enuresis after DVIU confirmed that bladder outlet obstruction was the cause of enuresis. However, the effect of DVIU was transient, as is often seen in adult urethral stricture. The American Urological Association guidelines for urethral stricture in adults recommend urethroplasty for recurrent bulbar stricture after undergoing an internal urethrotomy, although urethroplasty for pediatric enuresis is beyond their scope. We considered that urethroplasty should be deferred until completion of puberty, in the hope that urethral growth might improve the obstruction by increasing urethral caliber, or brain maturation might improve enuretic symptoms. When failure of the second DVIU was confirmed, puberty and body growth had been completed, and there was no longer any possibility that the stricture would resolve spontaneously, making urethroplasty a viable treatment choice.

Traditional bulbar urethroplasty by EPA has a risk of postoperative sexual problems, including de novo postoperative erectile dysfunction or painful ejaculation. NTU is a relatively novel procedure of bulbar urethroplasty with preservation of antegrade bulbar arterial flow. A multicenter study conducted in the United States reported that NTU is as effective as EPA for treating a non-traumatic stricture in adults, and has a lower risk of male sexual dysfunction. The stricture seen in this case could be classified as idiopathic and not associated with dense scarring as seen in traumatic strictures, and thus NTU was considered to be an optimal choice. Indeed, in the present case, enuresis was radically resolved after NTU in conjunction with improved urodynamic parameters without postoperative erectile dysfunction.

The generalizability of our case should be confirmed in future reports, although the success of our case does not mean that urethroplasty should be aggressively indicated as treatment for enuresis at a younger age because nocturnal enuresis is generally self-resolving. Also, NTU in the proximal bulbar urethra does pose a risk, albeit smaller than EPA, of sexual dysfunction, and may only be accepted by patients and parents after all other less invasive measures fail to resolve nocturnal enuresis.

In conclusion, an urethroplasty procedure, preferably an NTU upon completion of puberty, may be a good option for treating pediatric non-traumatic and short bulbar urethral stricture resistant to endoscopic treatment and causing refractory enuresis.

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**Author contributions**

Yuki Matsuo: Data curation; visualization; writing – original draft. Akihiro Kanematsu: Conceptualization; data curation; funding acquisition; methodology; project administration; writing – original draft; writing – review and editing. Shingo Yamamoto: Conceptualization; project administration; writing – review and editing.

**Conflict of interest**

The authors declare no conflict of interest.

**Approval of the research protocol by an Institutional Reviewer Board**

Not applicable.

**Informed consent**

Informed consent was obtained from the patient for publishing this case report.
Registry and the Registration No. of the study/trial

Not applicable.

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