



## Idiopathic urethral stricture in adolescents: A case series presentation and review of the literature

### Estenosis uretral idiopática en la adolescencia. Presentación de una serie de casos y revisión de la literatura

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#### Abstract

**Objective:** To present our case series of idiopathic urethral stricture in adolescents, analyzing the treatments applied and the results obtained.

**Methods:** A retrospective study of 7 patients with idiopathic urethral stricture treated at our center between 2000 and 2021 was performed.

**Results:** The average age at the moment of surgery was 14.7 years. In 85% of cases the main symptom was voiding difficulty. The diagnosis was by voiding cystourethrography (VCUG) in five patients and by cystoscopy in two, locating the stricture in the anterior urethra in all cases. Six patients were treated with endoscopic urethrotomy and one with balloon dilation. Two patients recurred. The subsequent treatment in both cases was a second endoscopic urethrotomy and one of them required a posterior urethroplasty. Currently 6 out of 7 patients are asymptomatic and the patient who underwent urethroplasty is undergoing regular dilations.

**Limitations:** The number of patients is limited due to the shortage of this pathology in childhood.

**Value:** The available literature is quite limited. It is usually secondary to trauma or surgical interventions such as hypospadias repair. Idiopathic etiology represents less than a third of urethral strictures in adolescents and it causes the greatest controversy regarding its management.

**Conclusions:** We should suspect idiopathic urethral stricture in childhood when symptoms appear, such as voiding difficulty in patients without previous pathologies. VCUG can help diagnosis, but a cystoscopy allows diagnosis and treatment at the same time. Endoscopic urethrotomy should be the treatment of choice for idiopathic strictures smaller than 1 cm, since the success rate is greater than 85%.

#### Keywords:

Idiopathic urethral stricture, adolescent, endoscopic urethrotomy

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## Resumen

**Objetivo:** Presentar nuestra serie de casos de estenosis uretral idiopática en adolescentes, analizando los tratamientos aplicados y los resultados obtenidos.

**Metodología:** Se realizó un estudio retrospectivo de 7 pacientes con estenosis uretral idiopática tratados en nuestro centro entre los años 2000 y 2021.

**Resultados:** La edad media en el momento de la cirugía fue de 14.7 años. En el 85% de casos el síntoma principal fue la dificultad miccional. El diagnóstico fue por cistouretrografía miccional seriada (CUMS) en cinco pacientes, y por cistoscopia en dos, localizando la estenosis en uretra anterior en todos ellos. Seis pacientes se trataron mediante uretrotomía endoscópica y uno mediante dilatación con balón. Recidivaron dos pacientes. El tratamiento posterior en ambos casos fue una segunda uretrotomía endoscópica y uno de ellos precisó una uretroplastia posterior. Actualmente 6 de los 7 pacientes están asintomáticos y el paciente de la uretroplastia se está realizando dilataciones periódicas.

**Limitaciones:** El número de pacientes es limitado debido a la escasez de esta patología en la infancia.

**Valor:** La literatura disponible es bastante limitada. Habitualmente es secundaria a traumatismos o a intervenciones quirúrgicas como la reparación de hipospadias. La etiología idiopática representa menos de un tercio de las estenosis uretrales en adolescentes y es la que mayor controversia presenta en cuanto a su manejo.

**Conclusiones:** Se debe sospechar estenosis uretral idiopática en adolescentes sin patologías previas si presentan síntomas como la dificultad miccional. La CUMS puede ayudar al diagnóstico pero una cistoscopia permite diagnosticar y tratar en el mismo acto. La uretrotomía endoscópica debe ser el tratamiento de elección ante estenosis idiopáticas menores de 1 cm, ya que la tasa de éxito es mayor del 85%.

### Palabras clave:

Estenosis uretral idiopática, adolescentes, uretrotomía endoscópica

## Background

Urethral stricture in childhood is considered a rare entity. The number of published series about urethral stricture is scarce, since most of them are about posterior urethral valves.<sup>(1-3)</sup> On the other hand, the literature on its management in children or adolescents is limited and most of the information is extrapolated from adults.

Urethral strictures are divided into two large groups according to their location in the anterior urethra (meatus, penile and bulbar urethra) and posterior urethra (membranous and prostatic urethra). Regarding the etiology, these

can have a traumatic, iatrogenic, inflammatory or idiopathic origin.<sup>(3,4)</sup> The group of idiopathic strictures is the one that causes the greatest controversy in terms of treatment due to its low frequency and its nature. There are very few series in the literature that focus on adolescent idiopathic strictures.

In the present study, a case series of adolescents with idiopathic urethral stricture is retrospectively assessed and key points of its diagnosis and treatment are analyzed.

## Materials and methods

A retrospective study was performed at the *Hospital Universitario 12 de Octubre* (Madrid, Spain), analyzing patients with urethral stricture treated during the period 2000-2021, aged between 10 and 17 years. Patients with strictures secondary to urethral surgery, congenital strictures, posterior urethral valves, or those secondary to trauma and inflammatory or infectious processes were excluded. A total of 7 cases were obtained in which the cause of the stricture was not found, being considered idiopathic. All patients included were male and had urinary symptoms. Uroflowmetry was performed in all of them, and voiding cystourethrography (VCUG) was performed in those in which obstruction parameters were obtained in uroflowmetry or when symptoms persisted despite having normal uroflowmetry. In all cases, the suspected diagnosis was confirmed by cystoscopy and the initial treatment was endoscopic.

Symptoms, diagnostic tests, location of the stricture, treatment performed and subsequent follow-up, as well as recurrences and salvage treatment were analyzed.

## Results

The average age at the time of surgery was 14.7 years (range 10-17). The most frequent symptom was voiding difficulty at the beginning of urination present in six patients (85.7%) followed by dysuria in two (28.5%), one of them having both symptoms. Patients with dysuria underwent urine culture, which was negative in all of them.

Ultrasound of the urinary system was normal in 100% of the cases and in two of them (28.5%) uroflowmetry showed a pattern of obstruction. A urethral stricture was identified in the VCUG in five patients (71.5%) and the rest was diagnosed by cystoscopy (28.5%). The cystoscopy showed normal bladder characteristics, with good-capacity, good-looking mucosa, normotopic ureteral orifices, without intravesical lesions in all of them. In all cases, the stricture was short (<1 cm) and located in the anterior urethra (one patient in the penile urethra (14.3%) and six in the bulbar urethra (85.7%)).

Endoscopic urethrotomy was performed in six cases (85.7%), with 10 Fr and 15,5 Fr Karl Storz Optical urethrotome depending of the penis's size, and balloon dilation in one (14.3%) with semicompliant balloon diameter of 7 mm (RX Muso Terumo), because an urethrotome was not available at that time.

All patients were evaluated with VCUG and uroflowmetry. In patients in whom the diagnosis was made by cystoscopy, cystoscopy was performed again 3 months after the procedure.

Two patients recurred (28.5%); one was after endoscopic urethrotomy (16.6% of those treated with this method) and the other one after balloon dilation (100% of those treated with this method). Subsequent treatment in both cases was a second endoscopic urethrotomy performed 6 and 10 months after the first one, respectively. One of them subsequently required an urethroplasty with resection of the stricture and primary anastomosis. This last patient has not achieved complete resolution of the stricture and is currently undergoing regular urethral dilations, initially monthly and thereafter every three months. The rest of the

patients (85.7%) currently have a good functional result after a mean follow-up of 5 years and 7 months (range 2-13 years). Data are collected in Table 1.

**Table 1. Summary of data of the case series**

Patient	Age (years)	Symptoms	Uroflowmetry	Diagnostic	Location	First treatment	Recurrence	Salvage treatment	Current functional outcome
1	16	Voiding difficulty	Normal	VCUG	Bulbar U.	Endoscopic urethrotomy	No	-	Good
2	14	Voiding difficulty	Altered	VCUG	Bulbar U.	Endoscopic urethrotomy	Yes	Urethrotomy + Urethroplasty	Urethral dilations
3	10	Voiding difficulty	Normal	VCUG	Bulbar U.	Endoscopic urethrotomy	No	-	Good
4	17	Dysuria	Normal	VCUG	Bulbar U.	Balloon dilation	Yes	Urethrotomy	Good
5	15	Dysuria + Voiding difficulty	Normal	Cistoscopy	Bulbar U.	Endoscopic urethrotomy	No	-	Good
6	15	Voiding difficulty	Normal	VCUG	Bulbar U.	Endoscopic urethrotomy	No	-	Good
7	16	Voiding difficulty	Altered	Cistoscopy	Penile U.	Endoscopic urethrotomy	No	-	Good

VCUG: Voiding cystourethrography; Bulbar U.: Bulbar urethra

## Discussion

The management of urethral strictures in pediatric population is a challenge since the number of reported cases is low and most conclusions are drawn from descriptive studies of short case series.

Regarding their location, they are grouped into strictures of the anterior urethra (meatus, penile and bulbar urethra) and posterior urethra (membranous and prostatic urethra). The recommended term is *stricture* when there is an abnormal narrowing of the anterior urethra with scarring and spongiofibrosis, and *stenosis* when it is located in the posterior urethra in the absence of spongiofibrosis.<sup>(1)</sup> In all the series consulted,<sup>(3,5-21)</sup> the anterior location was

much more frequent than the posterior one, as it was in our study. Ansari *et al.* reported the greatest series to date of urethral strictures in childhood with 195 cases, 141 (72.3%) in the anterior urethra and 54 (27%) in the posterior urethra.<sup>(3)</sup> In their study, most of them had a traumatic and iatrogenic origin, and 28.7% were idiopathic. In our series, 100% of the cases included were idiopathic, since the rest of the patients who had some associated urethral history were excluded.

There are authors who prefer to call them strictures of unknown etiology due to antecedents such as small traumas that may go unnoticed,<sup>(22)</sup> and they also speculate on the

possibility that they may be due to an alteration of embryogenesis with incomplete channeling of the urogenital membrane or to an anomaly Cowper's glands located in the floor of the bulbar urethra. However, the latter would be more in favour of strictures diagnosed in early childhood and not in adolescents, and it would be the theory of unnoticed trauma or urethritis with a later manifestation in adolescence that could be considered as a possibility in these cases.

One of the differences regarding posterior urethral valves (PUVs) is that in patients with urethral stricture, renal and bladder function is usually preserved, which suggests that the obstruction is less severe or acquired at a later time.<sup>(23)</sup> However, this difference is not always well recorded and it may actually be that cases of urethral stricture are underestimated, some being included as variants of PUV.<sup>(23)</sup>

Just like in our series, the most prevalent location is the bulbar urethra as it can be seen in other adult series.<sup>(24)</sup> Urinary symptoms reported by patients are varied and include dysuria, hematuria, urinary incontinence, voiding difficulty with increased emptying effort, and urinary retention.<sup>(2)</sup> In our study, the main associated symptom was voiding difficulty.

For diagnosis, there are different imaging tests and, as a non-invasive method, uroflowmetry can be useful. A weak flow or an enlarged voiding time would suggest an obstructive pattern and may lead to suspect this pathology. However, on many occasions this can be normal despite the presence of strictures.<sup>(1,2,25)</sup> and therefore it is useless to rule it out, a fact that is confirmed in our study since only a small proportion of patients had an altered uroflowmetry. To document stricture, the recommended tests by the Société Internationale d'Urologie (SIU) are VCUG, retrograde

urethrography and cystoscopy.<sup>(2)</sup> According to a retrospective study, urethral ultrasound can provide more precise data on the length and periurethral fibrosis in adolescents and can be useful as a complement to the rest of the tests for planning surgical intervention.<sup>(26)</sup>

For treatment of anterior urethral stricture, we can offer more conservative measures such as urethral dilation or urethrotomy under direct endoscopic vision, or more complex ones such as urethroplasty with resection and anastomosis or using grafts, with variable success and recurrence rates according to different published articles. The indications to carry out one or another technique are very controversial and proof of this is the disparity of opinion according to each author. In a recent review on the treatment of anterior urethral stricture in children, the cumulative success rate in 334 patients with endoscopic treatment by urethrotomy or dilation was 46% (with a range of 21%-75%), while the cumulative success rate in 347 patients after urethroplasty was 84% (with a range of 25%-100%).<sup>(27)</sup> There is a limitation in many studies regarding the definition of success when evaluating subjective symptoms instead of diagnostic tests.<sup>(18)</sup> In our series, in addition to the resolution of the symptoms, we performed an uroflowmetry, considered by some authors to be a good option to assess the functional result after treatment and lead to other tests in the event of an altered result.<sup>(9,27,28)</sup> Several groups agree that long-term endoscopic treatment is less effective and should not be used as the first option, since it often requires reintervention.<sup>(2,27,29)</sup>

Within the endoscopic treatment, urethrotomy is more accepted than urethral dilation, especially for short strictures less than 1 cm or as rescue of a short residual stricture after ure-

throplasty.<sup>(2,18,30)</sup> However, other authors defend endoscopic urethrotomy as initial treatment,<sup>(15)</sup> and according to our experience it seems to us to be a good option since, after a mean follow-up of 5 years and 7 months, only one patient has required reintervention. In adults, the most accepted curative treatment for bulbar urethral stricture is perineal urethroplasty and it can be considered as the first and definitive option in children,<sup>(1,27,31)</sup> or as salvage treatment after failure of a first urethrotomy.<sup>(1,15,18)</sup>

The main limitation of this study is that the number of cases analyzed is low and that it is a retrospective study, but we believe that due to the short number of published cases of idiopathic urethral stricture in adolescents, our data can contribute to the scientific literature to be able to reach a consensus for its best diagnostic and therapeutic management.

## Conclusions

Idiopathic urethral stricture is a rare entity in adolescent population, and we should suspect of it in those patients who have voiding difficulties without a previous urological history. A normal uroflowmetry does not rule out the presence of stenosis and we must request a VCUG to diagnose it, or perform a cystoscopy if the suspicion is high, since it allows an endoscopic treatment in the same act. Although there are few cases to draw conclusions, based on our experience and the literature we believe that endoscopic urethrotomy should be the treatment of choice for idiopathic strictures shorter than 1 cm located in the bulbar urethra. These patients require long-term follow-up due to their risk of recurrence, to whom repeated endoscopic treatments are not recommended

and the best option would be urethroplasty as definitive treatment. These conclusions should be interpreted with caution and multicenter studies with larger samples are needed to be able to develop treatment and follow-up protocols.

## CRedit Taxonomy

**Lara Herrero-López:** Term, Investigation, Visualization, Writing -Original Draft, Writing-Review and Editing.

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