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Congenital urethrocele in children. A case report

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ABSTRACT

INTRODUCTION: Congenital urethrocele is a rare disease in children. The diagnosis is often easy but the management remain difficult due to the risk of urethral stenosis form.

CASE PRESENTATION: We report a case of a 19-month-old child presenting with a penoscrotal mass. Cystourethroscopy confirmed the diagnosis of an urethrocele of the anterior urethra.

Urethrocele repair was performed with good results.

We propose to discuss clinical, paraclinical and therapeutic characteristics of congenital urethrocele in children.

CONCLUSION: An early, precise diagnosis and awareness of the anterior urethral diverticulum in boys with obstructive symptoms can reduce incidence of advanced uropathies.

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1. Introduction

Congenital urethrocele is a fairly rare disease in children [1,2]. It corresponds to a saccular dilatation of a portion of the urethral wall which may be less or more huge. The diagnosis is often easy but the management remain difficult due to the risk of urethral stenosis form. We report a case of a 19-month-old child presenting with a penoscrotal mass. We propose to discuss its clinical, paraclinical and therapeutic characteristics.

2. Case presentation

A 19-month-old boy with history of urinary tract infections, referred to our surgical department by family physician, for a poor urinary flow.

On examination he had a fluctuant swelling on the ventral face of the penile urethra (Fig. 1). This swelling was soft, cystic, fluctuant, and compressible. It collapsed completely on manual pressure with urine issue per urethra.

Urine analysis, routine blood counts and renal blood function were normal.

Ultrasonography showed normal kidneys in size and shape with a normal bladder. Cystourethroscopy confirmed an urethrocele of the anterior urethra (Fig. 2).

Surgical exploration found that the urethrocele was extending into the subcutaneous plane. Urethrocele repair was performed by excision and an urethroplasty using the Firlit technique which consists of a reconstruction of the mucous collar around the glans. This technique permits to obtain a normal appearance of a circumcised penis.

This procedure was performed by a paediatric surgeon with ten years of experience.

Post-operative recovery was uneventful. At 4 months follow-up the urinary stream was normal, with no urinary complaints. Parents are satisfied with the final result.

A clinical exam is planified once a year.

This work has been reported in line with SCARE 2018 criteria [13].

Consent to publish this case was obtained from the parent of study participant.

3. Discussion

Congenital anterior urethral diverticulum may be located all along the anterior urethra but it is usually located between the bulbous and the mid penile part. Urethral diverticulum is rarely situated in the distal urethra near the coronal sulcus.

Embryology of such urethral malformation remains unclear, but various hypotheses are proposed including a development defect of corpus spongiosum, cystic dilatation of the urethral glands, and sequestration of an epithelial nest after closure of the urethral folds. With a lack of a corpus spongiosum, a urethral dilatation in this region may develop into a diverticulum [3]

Abbreviations: VUR, vesicoureteral reflux.

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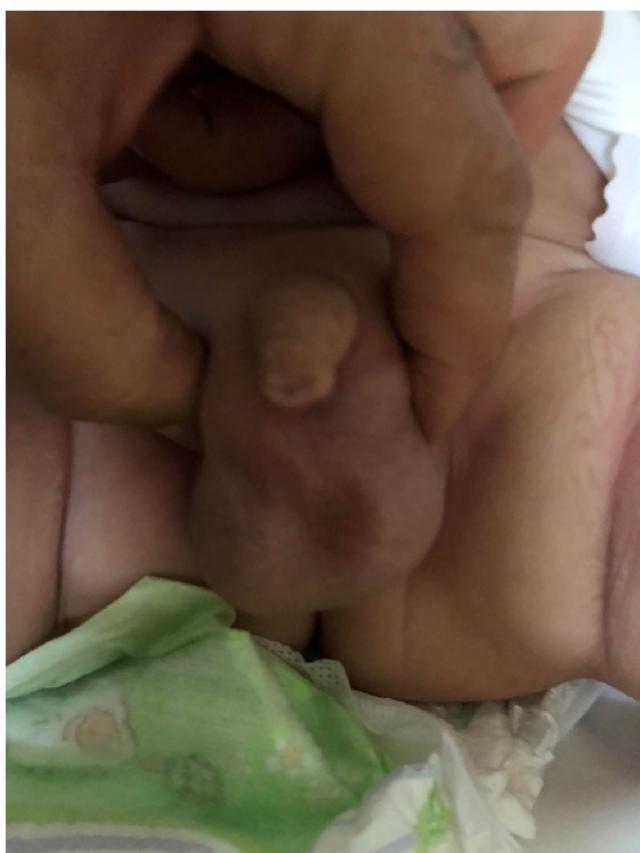


Fig. 1. Swelling on the ventral face of the penile urethra.



Fig. 2. Cystourethroscopy : urethrocele of the anterior urethra.

Clinical presentation depends essentially on the age and the degree of obstruction.

In the new born and infant, infectious symptoms (fever, diarrhoea, and vomiting) predominate and in older children, symptoms with voiding dominate (dysuria, thin urinary stream, frequency, urinary incontinence, and retention) [4]. During micturition a swelling may appear at penoscrotal region with post-void dribbling. Pressure on this swelling enables the emptying of its urinary contents.

Diagnosis rests essentially on the micturating cystourethrogram [2] which can also demonstrate proximal changes like megacystis, vesicoureteral reflux, or other associated anomaly. VUR has been reported in 20% of patients with anterior urethral diverticulum [5]

Endoscopy is usually difficult, particularly with the young child [6]. Ultrasonography is complimentary to evaluate the upper tract. Cystourethroscopy is diagnostic as well as therapeutic. A diverticulum typically appears as outpouching from the ventral wall of the urethra and has a proximal and distal rim [7].

The primary differential diagnostic conditions of anterior urethral diverticulum include anterior urethral valve, dilated Cowper's gland ducts, and post-traumatic diverticulum. The presence of a penile or penoscrotal mass clinically and the proximal lip radiologically which is seen as an arcuate filling defect should readily distinguish the diverticulum from the valve [8]. In dilated Cowper's gland ducts, a tubular channel is seen in the ventral surface of the bulbar urethra which it parallels, and its termination is in the urogenital diaphragm [4].

Management of the lesion can be by endoscopic or open surgery [2,9]. It depends on the size of the diverticulum and the degree of obstruction. Transurethral resection with a paediatric resectoscope is the treatment of choice for small diverticulum [7]. An open approach with patch graft urethroplasty can also be used to excise the diverticulum permanently and reconstruct the urethra, giving it a more uniform calibre. However, this option is associated to a high risk of urethrocutaneous fistula. We have associated an excision of diverticulum to a reconstruction by the Ferlit technique.

We can also find other technique like making a triangular flap which is fitted into the distal lip and double breasting of the urethral suture line, as described in literature [10].

Some authors have also advocated the placation of redundant diverticular wall with good results [11]. In situations where there are back-pressure changes of upper tracts with deranged renal function, urinary diversion [8] is a safer option.

Some authors advocate for asymptomatic forms that the treatment is based on the manual pressure of the urethra at the level of the diverticulum at the end of urination and the use of periodic antibiotic prophylaxis to prevent infections [12].

4. Conclusion

The diagnosis of anterior urethral diverticulum can be accurately made if this condition though less common is kept in mind. An early, precise diagnosis and awareness of the anterior urethral diverticulum in boys with obstructive symptoms can reduce incidence of advanced uropathies.

In patients of anterior urethral diverticulum with large diverticula without any back-pressure changes, as in the present paper, open diverticulectomy with primary repair is recommended as this procedure carries good results, and it takes care of the redundant diverticular wall.

Declaration of Competing Interest

No conflicts of interest

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Ethical approval

This study is exempt from ethical approval in our institution.

Consent

Consent to publish this case was obtained from the parent of study participant.

Author contribution

Study conception and design: Bchini

Acquisition of data: Ben Chouchen

Analysis and interpretation of data: Ben Chouchen

Drafting of manuscript: Ben Chouchen, Bchini, Ben Ahmed

Critical revision: Nouira, Jlidi

All authors provided critical feedback and helped shape the research, analysis and manuscript.

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References

- [1] M. Benjelloun, R. Rabii, H. Guessous, H. Essaki, S. Bennani, M. EL Mrini, Diverticule de l'urètre post-traumatique chez l'adulte, *Prog. Urol.* 13 (2003) 506–508.
- [2] P. Paulhac, L. Fourcade, N. Lesaux, J.L. Alain, P. Colombeau, Anterior urethral valves and diverticula, *BJU Int: EUUS series No 5* (2003) 506–509.
- [3] H.L. Kretschmer, Diverticula in the anterior urethra in male children, *J. Surgery Gynaecol. Obstetrics* 62 (1936) 634–640.
- [4] W.Y. Cheong, H.K. Cheng, K.P. Tan, Congenital anterior urethral diverticulum, *Singapore Med. J.* 29 (2) (1988) 171–175.
- [5] J.P. Murphy, K.W. Ashcraft, Anterior urethra diverticula and megalourethra, in: K.W. Ashcraft, T.M. Holder (Eds.), *Pediatric Surgery*, 2nd edition, Saunders, Philadelphia, Pa, USA, 1993, p. 643.
- [6] I.I. Karnak, M.E. Senocak, Rare congenital abnormalities of the anterior urethra, *Pediatr. Surg. Int.* 12 (1997) 407–409.
- [7] M. Zia-ul-Miraj, Congenital anterior urethral diverticula in children, *Pediatr. Surg. Int.* 15 (8) (1999) 567–569.
- [8] D.K. Gupta, M. Srinivas, Congenital anterior urethral diverticulum in children, *Pediatr. Surg. Int.* 16 (8) (2000) 565–568.
- [9] A. Kajbafzadeh, Congenital urethral anomalies in boys. Part II, *Urol. J.* 2 (3) (2005) 125–131.
- [10] R.Lal Bhatnagar, D.K. Mitra, Primary reconstruction of a congenital anterior urethral diverticulum, *Pediatr. Surg. Int.* 15 (3-4) (1999) 294–295.
- [11] B.W. Heaton, B.W. Snow, P.C. Cartwright, Repair of urethral diverticulum by plication, *Urology* 44 (5) (1994) 749–752.
- [12] Y. Sow, B. Sine, I.D. Diamé, N.A. Bagayogo, A. Thiam, D. Barboza, et al., Congenital Urethral Diverticulum in Male Subject: Report of Three Cases.
- [13] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A.J. Fowler, D.P. Orgill, et al., The SCARE 2018 statement: updating consensus Surgical CASE Report (SCARE) guidelines, *Int. J. Surg.* (2018).

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