Ejaculatory duct reflux revealed by chronic scrotal swelling in an adult

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ABSTRACT

Ejaculatory duct reflux is rare and few investigations have focused on this entity, which is usually described in children. This study reports a new case of unilateral ejaculatory reflux in a 32-year-old patient, with a history of urethroplasty at the age of 5 for hypospadias, who presented with right chronic scrotal swelling. Urethrocystoscopy showed an anterior urethral stricture and a gaping opening of the right ejaculatory duct. CT scan with opacification through the right ejaculatory duct, showed a dilated right seminal vesicle, associated with a reflux in the right deferent vas and epididymis, which was dilated explaining the scrotal swelling.

1. Introduction

Ejaculatory duct reflux (EDR) is rare and few investigations have focused on this entity, which is usually described in children and may have severe consequences on fertility. This study reports a case of unilateral ejaculatory reflux in an adult, revealed by chronic scrotal swelling.

2. Case presentation

A 32-year-old patient, with a history of scrotal skin graft urethroplasty at the age of 5 years for hypospadias, presented to our department with a right scrotal swelling that had been evolving for 2 years. On interrogation, the patient reported mild dysuria that had been present for years. Examination revealed an apical urethral meatus of normal caliber and a right scrotal swelling of 08 cm of great axis, painless on palpation. The digital rectal examination revealed a small prostate with a liquid-like mass adjacent to the prostate gland. The abdominal-pelvic CT scan showed a pelvic collection of 08 x 06 x 05 cm communicating through the inguinal canal with a right scrotal collection of 06 cm and a non-visualized right testicle. Given the clinical context, reflux of the right ejaculatory duct secondary to a urethral stricture was suspected, the pelvic collection would then correspond to the right seminal vesicle and the scrotal collection to the right epididymis. A retrograde opacification of the urethra could not be performed, due to the tortuous course of the urethra (the patient had a scrotal skin graft urethroplasty) with a catheter protruding only a few millimeters beyond the meatus. An urethrocystoscopy under anesthesia was then performed, finding a hairy urethra (testifying urethroplasty) and a stricture located at the junction between the urethroplasty and the native urethra, tight, which was opened with a cold knife (endoscopic internal urethrotomy). Exploration of the rest of the urethra revealed an opening of the right ejaculatory duct at the level of the verumontanum, which was gaping (Fig. 1). The opening of the left ejaculatory duct appeared normal. Intubation of the ejaculatory duct with a 7 French ureteral probe (Fig. 2) and opacification showed that it was indeed a dilated seminal vesicle. Cystoscopy showed a bladder with healthy mucosa and normal-appearing ureteral meatus. It was opted to keep the ureteral catheter in place with a bladder catheter. A pelvic CT scan was done immediately postoperatively, to confirm the diagnosis, and showed that the ureteral catheter was in the right seminal vesicle, which was opacified, associated to a reflux in the right deferent vas (Fig. 3). The ureteral catheter and the bladder catheter were removed 7 days after the operation and the patient started self-dilation. The patient was assessed at 3 months and showed good bladder voiding (no dysuria, no post-void residual). A pelvic MRI was performed, showing the disappearing of the dilatation of the right epididymis, and a decrease of the dilatation of the right seminal vesicle measuring 02 cm of great axis (Fig. 3).

3. Discussion

EDR is a rare entity that has been little described in the literature. Indeed, two mechanisms can be at the origin of the reflux. The first

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mechanism is an anatomical abnormality, due to a poorly defined angle of ejaculatory canal abutment to the prostatic urethra, a short submucosal course of the ejaculatory duct, or an ectopic opening. This mechanism has been suggested especially in children, and an association with other malformations is often found (anorectal malformation, hypospadias, Prune-Belly syndrome). The second mechanism is an obstacle to the flow of urine beyond the ejaculatory ducts, which has been described in both children (posterior urethral valve) and adults (urethral stricture). In these situations, the reflux is usually bilateral. In our case, a double mechanism can explain the reflux: The unilateral characteristic and the history of hypospadias surgery are in favor of an anatomical abnormality, associated with a narrowing of the urethra.

Reflux is difficult to diagnose, as the clinical presentation is not specific. It may be asymptomatic, or revealed by unexplained pelvic or scrotal pain, or recurrent genital infections (prostatitis, epididymitis, and orchitis). The consequences of EBU can be severe, resulting in infertility due to epididymal and testicular fibrosis secondary to infections and chronic inflammation. Our patient presented with a dilated epididymis, destroyed by the EBU with an atrophic testis. The diagnosis of EDR is confirmed radiologically, by opacification of the lower urinary tract, during retrograde urethrography, or at best by micturating cystourethrography. The reflux may be uni or bilateral and may involve the vas deferens, the seminal vesicle or the entire genital tract. In a retrospective study of EDR in the pediatric population including 54 cases, Wiersma found that reflux was limited to the seminal vesicle in 74% and that larger refluxes were associated with severe epididymal and testicular damage, such as the case described in this observation.

The treatment of EDR consists in correcting an underlying abnormality if it exists (urethral stricture, posterior urethral valves). Long-term antibiotic prophylaxis can also be proposed in case of recurrent genital infections, which can be difficult in children, with possible side effects. A vasectomy can be proposed in EDR with recurrent orchitis, to prevent pain and preserve the testicular parenchyma. The injection of a bulking agent into the verumontanum to prevent EDR has also been proposed by some authors. However, given the rarity of this condition, no treatment is currently recommended, except for the need to correct an underlying obstruction. In our case, the treatment of the urethral stricture allowed to decrease the importance of the reflux, as shown by the disappearance of the epididymal dilatation and the decrease of the seminal vesicle dilatation at the control imaging.

Fig. 1. Endoscopic view of the prostate urethra. a) Verumontanum (asterisk), gaping opening of the right ejaculatory duct (arrow). b) Intubation of the right ejaculatory duct by a guide. c) Placement of a ureteral probe in the right ejaculatory duct for opacification.

Fig. 2. Pelvic CT scan done after opacification through the catheter (placed in the right seminal vesicle). a) Dilated right seminal vesicle (red arrow); dilated vas deferens (blue arrow). b) Dilated right epididymis enhanced by the refluxing contrast (yellow arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)
4. Conclusion

EDR has a variable clinical presentation, with potentially serious consequences as far as infertility secondary to epididymal and testicular destruction. Underlying urethral obstruction must be treated. The management of malformative EDR is not well codified, and vasectomy or injection may be proposed in severe cases.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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Declaration of competing interest

The authors declare that there are no conflicts of interest regarding the publication of this article.

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Fig. 3. Pelvic MRI control 03 months after surgery (T2 sequence). a) Sagittal section showing the regression of the dilatation of the right seminal vesicle (blue arrow). The red arrow shows the bladder. b) Axial section: Disappearance of the dilatation of the right vas deferens (yellow arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)