Submitted: 19 July, 2024 Accepted: 24 October, 2024 Published: 30 March, 2025 DOI:10.22514/j.androl.2025.012

REV INT ANDROL
Revista Internacional de Andrología

CASE REPORT

Urethrocavernous fistula: a case report and systematic review

Javier Fernández Siles[1,*], Sergio Correa Portillo[1,*], Rodrigo España Navarro[1,*]

[1]Urology Department, Hospital Regional Universitario de Málaga, 29009 Málaga, Spain

***Correspondence**
javier.fernandez.sspa@juntaandalucia.es (Javier Fernández Siles);
sergio.correa.sspa@juntaandalucia.es (Sergio Correa Portillo);
rodrigo.espana.sspa@juntaandalucia.es (Rodrigo España Navarro)

Abstract

Background: Urethrocavernous fistulas are rare pathological communications between the urethra and corpora cavernosa, primarily associated with trauma or iatrogenic interventions (*e.g.*, priapism shunt surgery). Their nonspecific symptomatology (urethrorrhagia, purulent discharge, or urinary retention) often delays diagnosis. The objective of this paper is to analyze the clinical and therapeutic profile of urethrocavernous fistulas through a case report and systematic literature review to establish evidence-based management insights. **Case**: A 22-year-old male presented with recurrent urethrorrhagia during erection and acute urinary retention secondary to clots. The initial urethroscopy and CT angiography were inconclusive. Subsequent arteriography revealed a right internal pudendal artery-cavernosal fistula. After two non-permanent embolizations failed, definitive occlusion was achieved via non-absorbable coils, resolving symptoms without recurrence or other complications at 23 months. **Conclusions**: Urethrocavernous fistulas are rare with limited literature descriptions. Systematic review of 17 cases reveals marked heterogeneity in the clinical presentations and the therapeutic outcomes, emphasizing the imperative for standardized diagnostic and therapeutic protocols. They are predominantly diagnosed incidentally through retrograde urethrography and urethroscopy. Conservative management with urinary diversion may be considered as the initial approach, while surgery is reserved for refractory cases or those with associated injuries. Embolization is a poorly studied treatment option.

Keywords

Urethrocavernous fistula; Retrograde urethrography; Trauma; Surgery; Conservative management

Fístula uretrocavernosa: un reporte de un caso y una revisión sistemática

Resumen

Antecedentes: Las fístulas uretrocavernosas son comunicaciones patológicas infrecuentes entre la uretra y los cuerpos cavernosos, asociadas principalmente a traumatismos o intervenciones iatrogénicas (ej. cirugía de derivación para priapismo). Su sintomatología inespecífica (uretrorragia, secreción purulenta o retención urinaria) suele retrasar el diagnóstico. El objetivo de este artículo es analizar el perfil clínico-terapéutico de estas fistulas mediante un reporte de caso y una revisión sistemática de la literatura, con el fin de establecer pautas de manejo basadas en evidencia. **Caso**: Varón de 22 años presentó uretrorragia recurrente durante la erección y retención urinaria aguda secundaria a coágulos. La uretroscopia y la angiografía por tomografía computarizada (TC) iniciales fueron inconclusas. Una arteriografía posterior identificó una fístula entre la arteria pudenda interna derecha y el cuerpo cavernoso ipsilateral. Tras el fracaso de las dos embolizaciones temporales con material absorbible, se logró la oclusión definitiva mediante coils no absorbibles, resolviendo los síntomas sin recurrencia ni complicaciones a los 23 meses. **Conclusiones**: Las fístulas uretrocavernosas son entidades raras con escasa descripción en la literatura. La revisión sistemática de 17 casos reveló una heterogeneidad destacable en sus presentaciones clínicas y resultados terapéuticos, subrayando la necesidad de protocolos diagnósticos y terapéuticos estandarizados. Su diagnóstico es predominantemente incidental mediante uretrografía retrógrada y uretroscopia. El manejo conservador con derivación urinaria puede considerarse como abordaje inicial, reservándose la cirugía para casos refractarios o con lesiones asociadas. La embolización constituye una opción terapéutica poco estudiada.

Palabras Clave

Fístula uretrocavernosa; Uretrografía retrógrada; Traumatismo; Cirugía; Tratamiento conservador

1. Introduction

Urethrocavernous fistulas are rare clinical entities characterized by abnormal communications between the urethra and the corpora cavernosa. These fistulas may develop as a consequence of trauma, infections or as complications of surgeries, such as proximal shunt procedures for refractory priapism or penile prosthesis implantation. The existing literature on urethrocavernous fistulas is limited, with reported cases exhibiting a heterogeneous clinical spectrum with inconsistent signs like urethrorrhagia, purulent discharge, or erectile dysfunction. Due to their infrequent occurrence, urethrocavernous fistulas are often diagnosed incidentally, primarily through retrograde urethrography. Currently, there is no standardized approach to management, with both conservative and surgical treatments yielding varied outcomes. We present a case of a urethrocavernous fistula manifesting as urethrorrhagia during erection, which was resolved after three embolizations. Subsequently, we conducted a systematic review of the existing literature to provide a comprehensive analysis of this condition.

2. Clinical case

A 22-year-old male presented to the emergency department of our hospital with urethrorrhagia during erections, which resolved spontaneously, and urinary difficulty due to the presence of clots, with no other associated symptoms. He reported three similar episodes in the preceding week, with no history of trauma, high-risk sexual behavior, urinary tract infection, lithiasis, voiding symptoms or fever. The patient did not experience priapism and his sexual function remained unaffected. On physical examination, he exhibited bladder distension and tenderness upon palpation of the hypogastrium, without signs of peritoneal irritation. Acute bladder retention secondary to clot formation was diagnosed, requiring bladder catheterization and continuous bladder irrigation. After twelve hours of irrigation, the urine cleared, and the patient was discharged with successful removal of the urinary catheter. Laboratory tests did not show any significant abnormalities, with a hemoglobin of 14 g/dL (13–17 g/dL) and a creatinine level of 1.04 mg/dL (0.7–1.3 mg/dL).

After twelve days at home, the patient was readmitted to the Urology department with recurrent urethrorrhagia and acute urinary retention. His laboratory results showed a hemoglobin of 15 g/dL (13–17 g/dL) and a creatinine level of 1 mg/dL (0.7–1.3 mg/dL). Urinary catheterization and continuous bladder irrigation were required again, and the urethrorrhagia resolved within 48 hours. Diagnostic evaluations, including urethroscopy, retrograde urethrography, Doppler ultrasound, and CT angiography, revealed no pathological findings. After a three-day hospital stay, the patient was discharged without a urinary catheter and clear urine.

Seven weeks later, an arteriography identified a small fistula between the right internal pudendal artery and the right corpus cavernosum. A non-permanent embolization using Spongostan® was performed (Fig. 1), and a repeat embolization was required seven months later due to persistent symptoms (Fig. 2), following two additional episodes of urethrorrhagia that were treated in the emergency department. Five months after the second intervention, urethrorrhagia recurred, necessitating two more emergency department visits. This prompted a selective embolization with non-absorbable coils, targeting the branches responsible for the fistula between the bulbourethral artery and cavernous artery, while preserving the dorsal penile artery (Fig. 3). This procedure successfully resolved the fistula.

Throughout follow-up, the patient's hemoglobin level did not decrease significantly, and did not reach anemic range during any episode. Additionally, there were no signs of renal failure. The patient remains asymptomatic at the 23-month follow-up since the initial episode.

3. Materials and methods

A case of urethrocavernous fistula has been reported. We conducted a systematic review in accordance with PRISMA guidelines [1]. A comprehensive search was performed in the Scopus, PubMed, Cochrane and Embase databases, using the following query: ((urethro-cavernosal) or (urethrocavernous) or (urethra and cavernous)) and (fistula or fistulae). All records up to 2023 were considered. The flowchart is shown in Fig. 4. Data extracted from the reviewed cases included patient age, clinical presentation, diagnosis method, fistula size and location, treatment modality, causative mechanism, time to symptom onset after the causative event, follow-up duration and post-treatment complications.

4. Results

We analyzed 17 cases: 14 were identified through the database research, and three were found through bibliographic review. The average patient age was 44 years. The most common clinical manifestations were urethral purulent discharge (n = 6) and urethrorrhagia (n = 6). Retrograde urethrography was the predominant diagnostic modality (n = 8). The most frequently affected urethral region was the bulbar area (n = 6), and the right corpus cavernosum was the most commonly involved (n = 6). Trauma was the predominant causative mechanism (n = 8), followed by shunt surgery for priapism treatment (n = 5). The mean time to symptom onset after the causative event was 46 days. Surgical treatment was the most common approach (n = 8), with direct fistula repair being the prevailing method (n = 6). The average follow-up duration was 249 days, with 8 cases experiencing complications, including urethral diverticulum (n = 3) and erectile dysfunction (n = 3). Findings are summarized in **Supplementary Table 1**.

5. Discussion

Urethrocavernous fistulas are rare urological conditions with limited documented cases in the literature. Clinical manifestations such as urethral purulent discharge [2–7] and urethrorrhagia [8–13] are nonspecific and can mimic signs of more common urological conditions such as urethritis [14] and urethral trauma [15]. Diagnosis is primarily achieved through retrograde urethrography [2–5, 8, 11, 16, 17], a widely used test in urological evaluations. Retrograde urethrography is the preferred modality for initial assessment of urethral trauma,

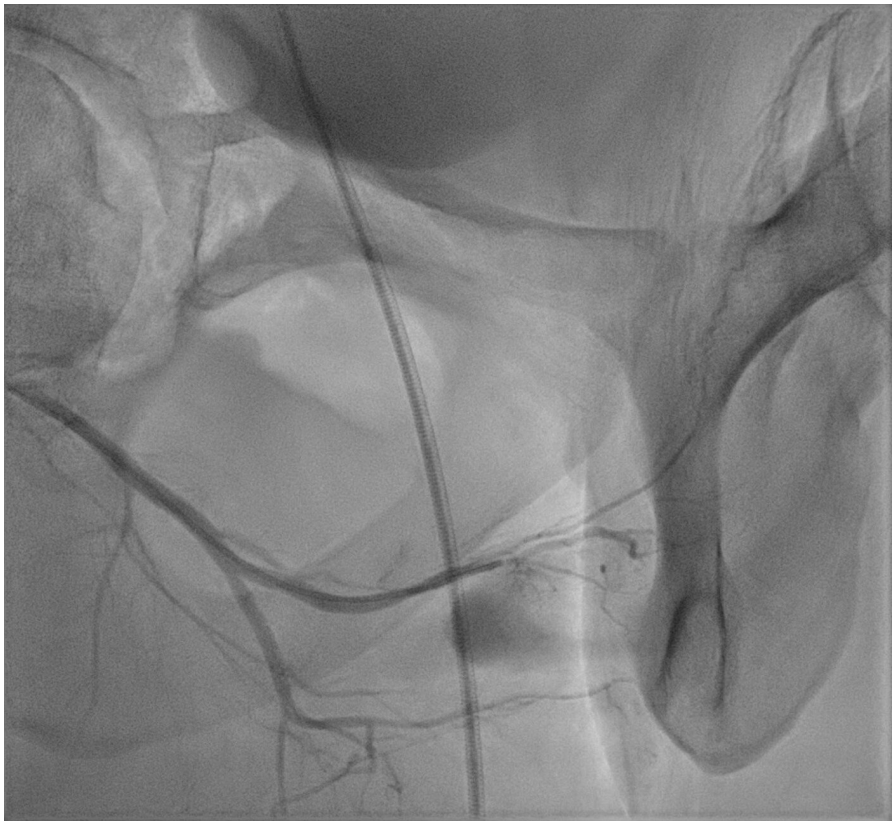

FIGURE 1. Initial non-permanent embolization with Spongostan® of fistula between the right internal pudendal artery and the right corpus cavernosum.

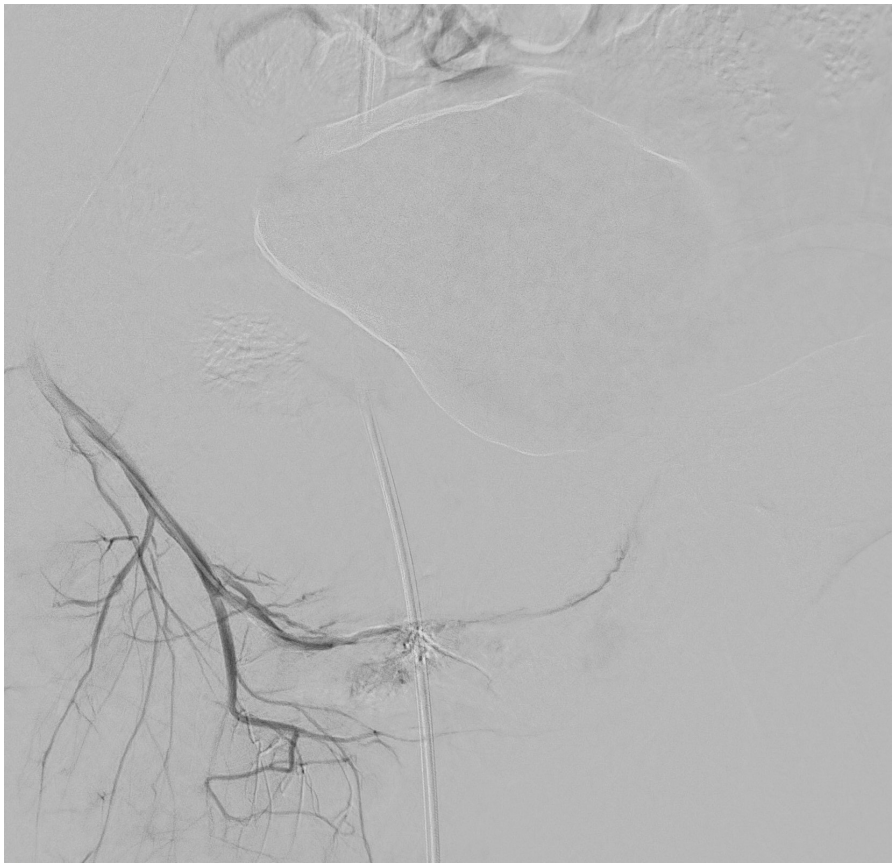

FIGURE 2. Second non-permanent embolization with Spongostan® of the same fistula.

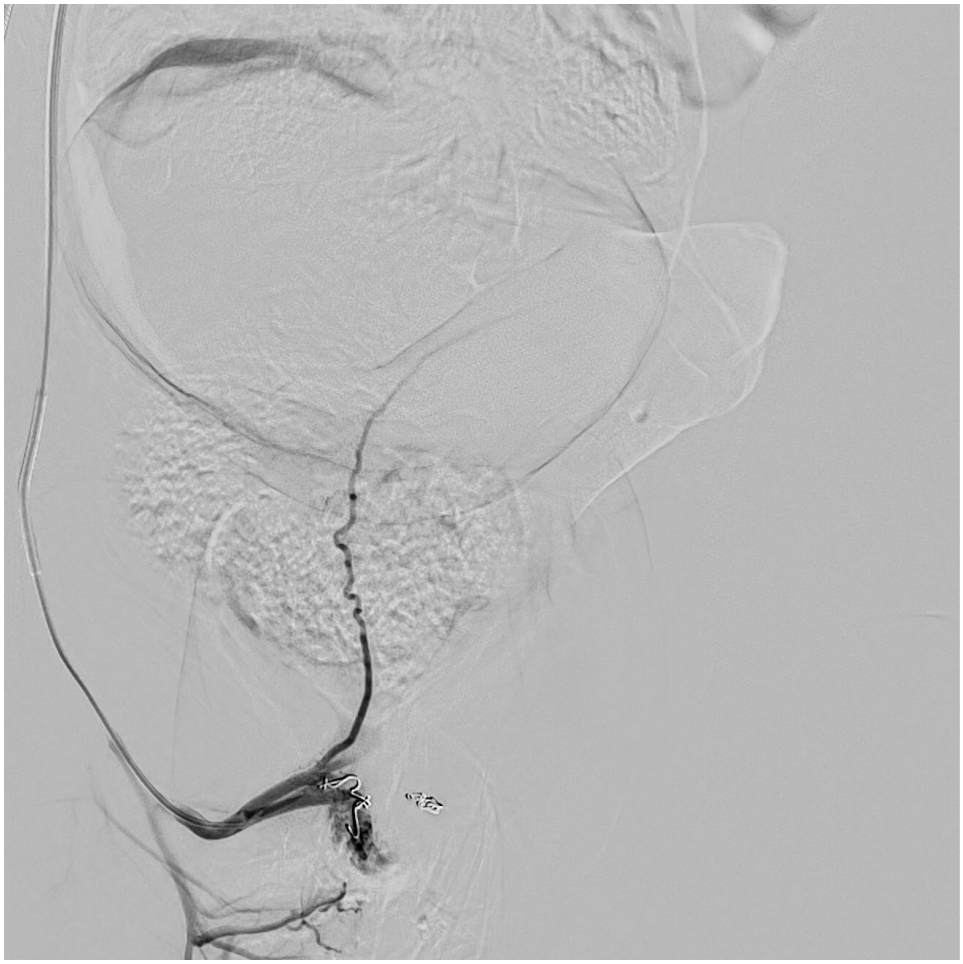

FIGURE 3. Embolization with non-absorbable coils.

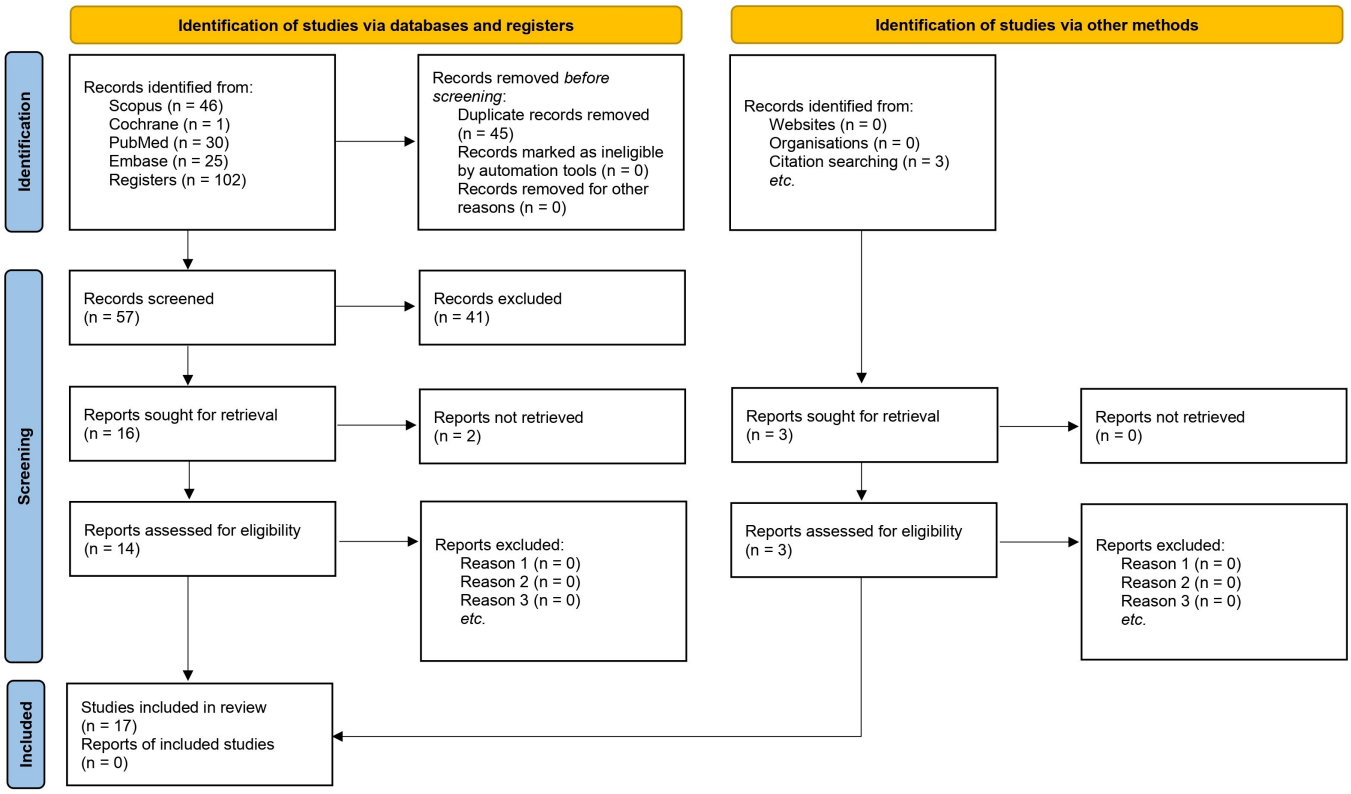

FIGURE 4. Flowchart based on PRISMA guidelines.

although cystourethroscopy is also a valid alternative [15]. The existing literature does not provide a clear explanation for the higher frequency of involvement in the bulbar urethra [6–8, 10, 13, 18] and the right corpus cavernosum [2, 3, 8–11]. Identified causative mechanisms include physical trauma [8–11, 13, 16, 19, 20], shunt surgery for priapism treatment [2–5, 17], penile prosthesis replacement [7, 18], and urethrotomy with intralesional mitomycin injection for urethral stricture surgery [6]. In industrialized countries, iatrogenesis is the leading cause of urinary fistulas [21]. Following the causative event, fistulas manifested several weeks later, suggesting a need for *post hoc* suspicion, although larger sample studies are required for confirmation.

Surgical management is recommended for patients refractory to or predicted to fail conservative management [22]. For urethral trauma, immediate reconstruction is indicated for complete ruptures and those associated with penile fractures, but it is contraindicated initially in unstable patients or those with pelvic fractures [15]. Due to the limited literature on urethrocavernous fistulas, there is no robust evidence for specific predictors guiding treatment decisions. Management decisions are based on accumulated experience from urethral trauma, urinary fistulas, hypospadias and urethroplasties [15, 22]. Two cases employed fibrin sealant patches (Tachosil®, Takeda Austria GmbH, Linz, Austria) [18] and pericardial allografts (Tutoplast®) [12] for fistula repair, both resulting in complete remission. Tachosil® has been used in rectourethral fistulas [23] and vesicovaginal fistulas [24]. Tutoplast® has been used as graft material for tunica albuginea in Peyronie's disease, with documented applications in testicular and bladder reconstructions as well [25]. Further studies are needed to assess the efficacy of these materials in urethrocavernous fistula repairs.

Conservative management principally involves urinary diversion with suprapubic catheter [2–5, 11, 16, 17], with one case managed with urethral catheterization [6]. For anterior urethral trauma in males, there is no evidence supporting the superiority of suprapubic over transurethral catheters with endoscopic realignment [15]. The recommended duration of urinary diversion is one-two weeks for partial ruptures and three weeks for complete ruptures [26, 27]. Due to case heterogeneity and the small sample size, there is no clear evidence on the superiority of one method over another. Antibiotics are often included in conservative management [5, 6, 11, 19] to address concurrent urinary infections.

Urethral diverticula occurred in cases related to proximal shunt surgery for priapism, initially managed conservatively with urinary diversion [2, 4, 5]. Erectile dysfunction was observed in cases that underwent proximal shunt surgery for priapism [3, 5]. In the cases with fistula resulting from proximal shunt surgery for priapism (n = 5), complications were noted in all but one case, which had no post-surgical follow-up. Proximal shunt surgery is more invasive and associated with a higher complication rate compared to distal shunt surgery. The American Urological Association (AUA) and European Association of Urology (EAU) guidelines recommend distal shunt procedures prior to considering proximal shunt surgery [28, 29]. Further research is needed to evaluate complications associated with urethrocavernous fistulas.

Our case presented with urethrorrhagia during erections, an unusual manifestation in the reviewed literature [8, 10]. Neither arteriography nor embolization for fistula management is documented in the selected cases. Persistent urethrorrhagia and urinary retention are also rare in the literature, with only one article reporting such complications [12]. Embolization is a technique with a low complication rate, although recent literature has reported glans necrosis as a complication of prostatic artery embolization [30, 31].

This study has limitations. The reviewed clinical cases were not collected following a common standardized approach, resulting in heterogeneity and incomplete information. The small number of cases further limits the conclusions. Future studies should aim for homogeneous data and compare treatment options to enhance understanding of this condition. Improved knowledge could facilitate better diagnosis and treatment.

6. Conclusions

Urethrocavernous fistulas are rare urological pathologies that present with nonspecific symptoms and can be diagnosed through retrograde urethrography and/or cystourethroscopy. The causative mechanisms often involve physical agents, particularly trauma and iatrogenesis. Management is based on experience derived from other urological conditions, due to the lack of specific studies on this entity. Conservative treatment with urinary diversion should be considered as the initial approach, reserving surgery for complete urethral ruptures, penile fractures, conservative management failure and subsequent interventions. The role of embolization as a treatment remains uncertain and requires further investigation to determine its viability as a therapeutic option.

AVAILABILITY OF DATA AND MATERIALS

The data and materials used in this case report and review are contained in this article.

AUTHOR CONTRIBUTIONS

JFS—collected the clinical case data, performed the systematic review, and wrote the initial version of the manuscript. SCP—performed the systematic review and revised the correct expression and terminology of the article. REN—designed the study and reviewed the methodology. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The research was conducted in accordance with the guidelines of the Declaration of Helsinki (as revised in 2024). Written informed consent was obtained from the patient for the publication of anonymized information in this article. The Hospital Regional Universitario de Málaga does not require ethical approval for reporting individual cases or case series.

ACKNOWLEDGMENT

We would like to thank our colleagues from the Urology Department at our hospital for his assistance and guidance in this research.

FUNDING

This research received no external funding.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

SUPPLEMENTARY MATERIAL

Supplementary material associated with this article can be found, in the online version, at https://files.intandro.com/files/article/1905452362763255808/attachment/Supplementary%20material.docx.

REFERENCES

[1] Page MJ, Moher D, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, *et al*. PRISMA 2020 explanation and elaboration: updated guidance and exemplars for reporting systematic reviews. The BMJ. 2021; 372: n160.

[2] Klugo RC, Olsson CA. Urethrocavernous fistula: complication of cavernospongiosal shunt. The Journal of Urology. 1972; 108: 750–751.

[3] Buckspan M, Klotz P. Urethrocavernous fistula: a case report. The Journal of Urology. 1977; 117: 538.

[4] Robbins DM, Crawford ED, Lackner HL. Late development of a urethrocavernous fistula after cavernospongiosum shunt for priapism. The Journal of Urology. 1984; 132: 126–127.

[5] Meyer J-P, Gingell C, Dickerson D. An unusual complication of a spongio-cavernosal shunt. BJU International. 2002; 90: e36–e37.

[6] Rehder P, Pedrini M, Jelisejevas LA, Gulacsi A, Horninger W, Stuehmeier J. Urethro-cavernosal-spongiosal fistula after intralesional mitomycin C for recurrent urethral stricture disease. Urology Case Reports. 2020; 33: 101281.

[7] Botkin H, Barnes B, Pearlman A. Bulbar urethrocavernous fistula in setting of inflatable penile prosthesis: a case report. BMC Urology. 2021; 21: 25.

[8] Ochsner MG, Joshi PN. Urethrocavernosus fistula. The Journal of Urology. 1982; 127: 1190.

[9] Hargreaves DG, Plail RO. Fracture of the penis causing a corporo-urethral fistula. British Journal of Urology. 1994; 73: 97.

[10] Seftel AD, Matthews LA, Herbener TE, Spirnak JP. Corpus cavernosum-spongiosum fistula after blunt pelvic trauma: successful resolution with digoxin. The Journal of Urology. 1996; 156: 1769.

[11] Juaneda Castell B, Montlleó González M, Ponce de León Roca X, Gausa Gascón L, Caparrós Sariol J, Villavicencio Mavrich H. Urethrocavernous fistula due to penile fracture. Actas Urológicas Españolas. 2008; 32: 1043–1045. (In Spanish)

[12] Pearlman AM, Rukstalis DB, Terlecki RP. Diagnosis and treatment of urethrocavernous fistula presenting as urethral bleeding. Urology. 2018; 120: e5.

[13] Han J, Ghodoussipour S, Doumanian LR. Delayed presentation of urethro-cavernosal fistula after urotrauma. Urology. 2019; 131: e7–e8.

[14] Bonkat G, Bartoletti R, Bruyère F, Cai T, Geerlings SE, Köves B, *et al*. EAU guidelines on urological infections. European Association of Urology: Amsterdam, Netherlands. 2022.

[15] Kitrey ND, Campos-Juanatey F, Hallscheidt P, Mayer E, Serafetinidis E, Sharma DM, *et al*. EAU guidelines on urological trauma. EAU Guidelines Office: Arnhem, Netherlands. 2023.

[16] Palaniswamy R, Rao MS, Bapna BC, Chary KS. Urethro-cavernous fistula from blunt penile trauma. The Journal of Trauma. 1981; 21: 242–243.

[17] Manjunath AS, Mazur DJ, Han JS, Gonzalez CM. Simultaneous urethro-cutaneous and urethrocavernous fistula after proximal corporospongiosal shunt for priapism. Urology. 2015; 85: e13–e14.

[18] Caraceni E, Marronaro A, Leone L. Salvage procedure in case of urethrocavernous fistula after revision surgery for malfunctioning three-piece penile prosthesis. Case Reports in Urology. 2016; 2016: 4179862.

[19] Motiwala HG. Urethrocavernous fistula following sexual intercourse. The Journal of Urology. 1993; 149: 371.

[20] Capretti C, Avolio A, Florio A, Giovannozzi S, de Carolis D. Management of complications in penile prosthesis reimplantation: a case report. Archivio Italiano di Urologia e Andrologia. 2020; 92: 215–216.

[21] De Ridder D, Greenwell T. Urinary track fistulae. In Partin AW, Dmochowski RR, Kavoussi LR, Peters CA, Wein AJ (eds.) Campbell-Walsh-Wein urology (pp. 2924–2963). 12th edn. Elsevier: Philadelphia. 2021.

[22] Han J, Ghodoussipour S, Kreydin EI, Doumanian L. Diagnosis and management of urethro-cavernosal fistulas. Journal of Clinical Urology. 2020; 13: 398–401.

[23] Giuliani G, Guerra F, Coletta D, La Torre M, Franco G, Leonardo C, *et al*. Repair of transperineal recto-urethral fistula using a fibrin sealant haemostatic patch. Colorectal Disease. 2016; 18: O432–O435.

[24] Giusti G, Lucci Chiarissi M, Abate D, De Vita G, Angioni S, De Lisa A. Early repair of post-hysterectomy vesicovaginal fistulae through a laparoscopic transperitoneal extravesical approach. Experience of a Single Center. Urology. 2018; 119: 44–48.

[25] Hosseini J, Hosseini S, Hosseini MA, Rezaei Y. Pericardium in reconstructive urologic surgeries: a systematic review and meta-analysis. Urologia Internationalis. 2019; 102: 131–144.

[26] Elgammal MAA. Straddle injuries to the bulbar urethra: management and outcome in 53 patients. International Brazilian Journal of Urology. 2009; 35: 450–455.

[27] Brandes S. Initial management of anterior and posterior urethral injuries. Urologic Clinics of North America. 2006; 33: 87–95.

[28] Bivalacqua TJ, Allen BK, Brock GB, Broderick GA, Chou R, Kohler TS, *et al*. The diagnosis and management of recurrent ischemic priapism, priapism in sickle cell patients, and non-ischemic priapism: an UA/SMSNA guideline. The Journal of Urology. 2022; 208: 43–52.

[29] Salonia J, Bettocchi C, Capogrosso P, Carbalho J, Corona G, Hatzichristodoulou G, *et al*. EAU guidelines on sexual and reproductive health. European Association of Urology: Amsterdam. 2024.

[30] Chung E. Penile glans necrosis following prostatic artery embolization for benign prostatic hyperplasia: case series and review of current literature. World Journal of Men's Health. 2023; 41: 396–402.

[31] Mouli S, Salem R, McClure TD. Prostate artery embolization for benign prostatic hyperplasia. The Journal of Urology. 2024; 212: 216–219.

How to cite this article: Javier Fernández Siles, Sergio Correa Portillo, Rodrigo España Navarro. Urethrocavernous fistula: a case report and systematic review. Revista Internacional de Andrología. 2025; 23(1): 102-107. doi: 10.22514/j.androl.2025.012.