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CASE REPORT

Urethrocavernous fistula: a case report and systematic review

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Abstract

Background: Urethrocavernous fistulas are rare conditions that are often diagnosed incidentally, characterized by abnormal communications between the urethra and the corpora cavernosa. The objective of this paper is to review the existing literature on urethral cavernous fistulas based on the description of a clinical case. **Methods**: A case of urethrocavernous fistula in our clinical setting is presented and a systematic review was conducted using Scopus, PubMed, Cochrane, and Embase databases. **Results**: Seventeen cases were analyzed. The most common symptoms were urethral purulent discharge (n = 6) and urethrorrhagia (n = 6). Retrograde urethrography was the most commonly employed diagnostic modality (n = 8). Trauma was the predominant causative mechanism (n = 8), followed by shunt surgery for priapism treatment (n = 5). Surgical treatment was the most common approach (n = 8), with immediate fistula repair being the most prevalent method (n = 6). **Conclusions**: Urethrocavernous fistulas are rare with limited literature descriptions. They are predominantly diagnosed incidentally through retrograde urethrography and cystourethroscopy. 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 entidades infrecuentes que se diagnostican a menudo de forma incidental, y que se caracterizan por comunicaciones anómalas entre la uretra y los cuerpos cavernosos. El objetivo de este artículo es revisar la literatura existente sobre las fístulas uretrocavernosas a partir de la descripción de un caso clínico. **Métodos**: Se presenta un caso de fístula uretrocavernosa en nuestro medio y se realizó una revisión sistemática utilizando las bases de datos Scopus, PubMed, Cochrane y Embase. **Resultados**: Se analizaron 17 casos. Los síntomas más frecuentes fueron secreción uretral purulenta (n = 6) y uretrorragia (n = 6). La uretrografía retrógrada fue la prueba diagnóstica más frecuente (n = 8). El traumatismo fue el mecanismo causal predominante (n = 8), seguido de la cirugía tipo shunt para el tratamiento del priapismo (n = 5). El tratamiento quirúrgico fue el manejo más frecuente (n = 8), siendo la reparación inmediata de la fístula la opción más prevalente (n = 6). **Conclusiones**: Las fístulas uretrocavernosas son entidades infrecuentes con escasas descripciones en la literatura. Se diagnostican predominantemente de forma incidental mediante uretrografía retrógrada y cistouretroscopia. El tratamiento conservador con derivación urinaria puede considerarse la medida terapéutica inicial, mientras que la cirugía se reservaría para casos refractarios o con lesiones asociadas. La embolización es 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,

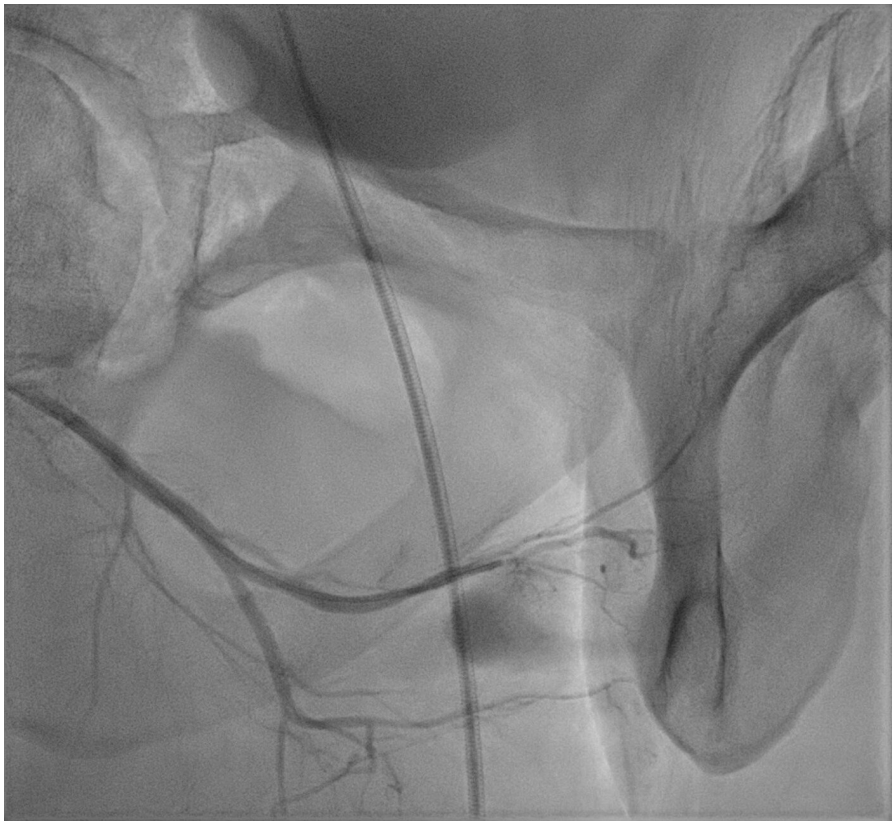

FIGURE 1. Initial non-permanent embolization with Spongostan® of fistula between the right internal pudendal artery and the right corpus cavernosum.

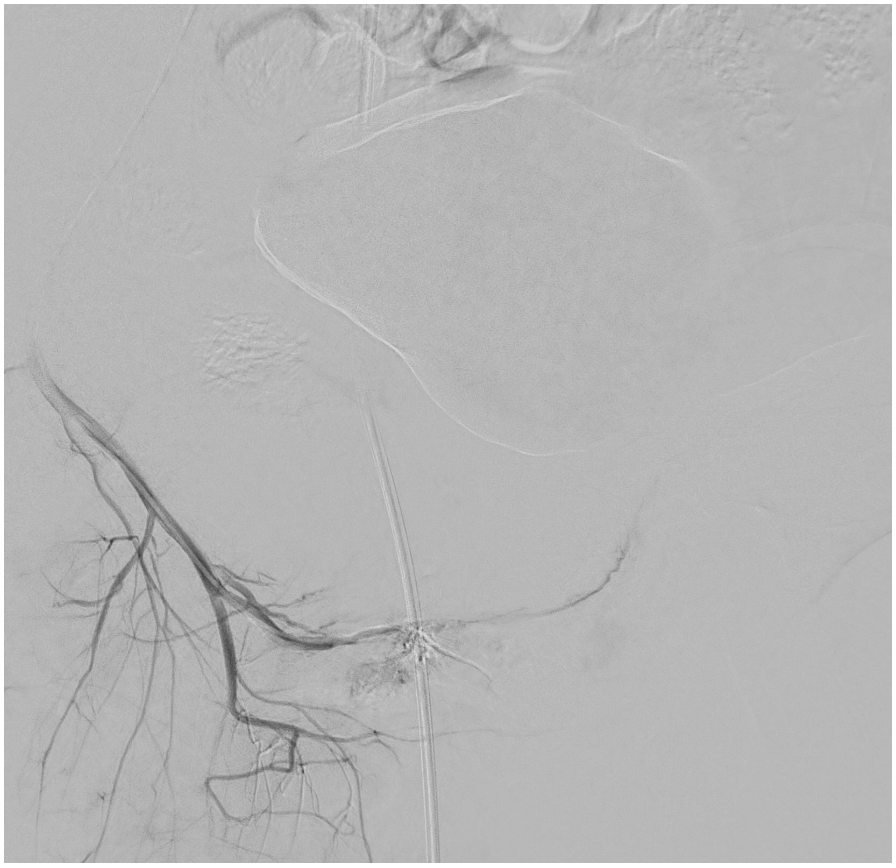

FIGURE 2. Second non-permanent embolization with Spongostan® of the same fistula.

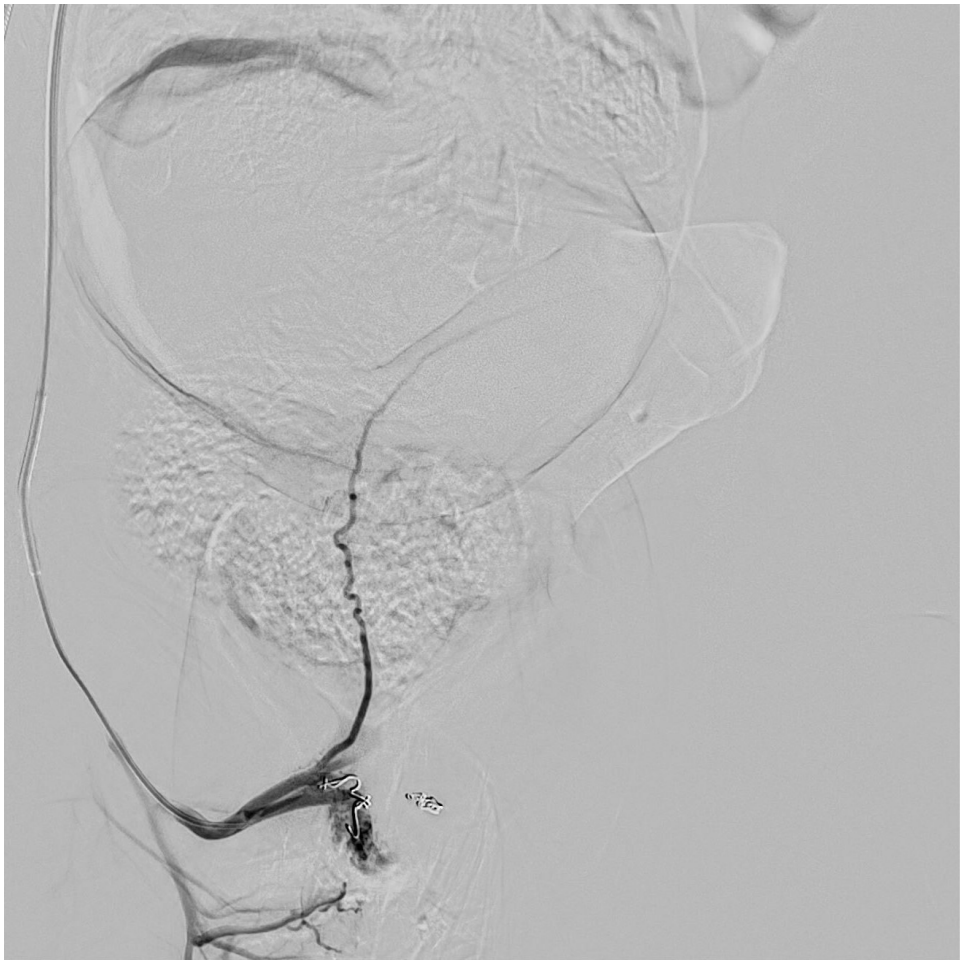

FIGURE 3. Embolization with non-absorbable coils.

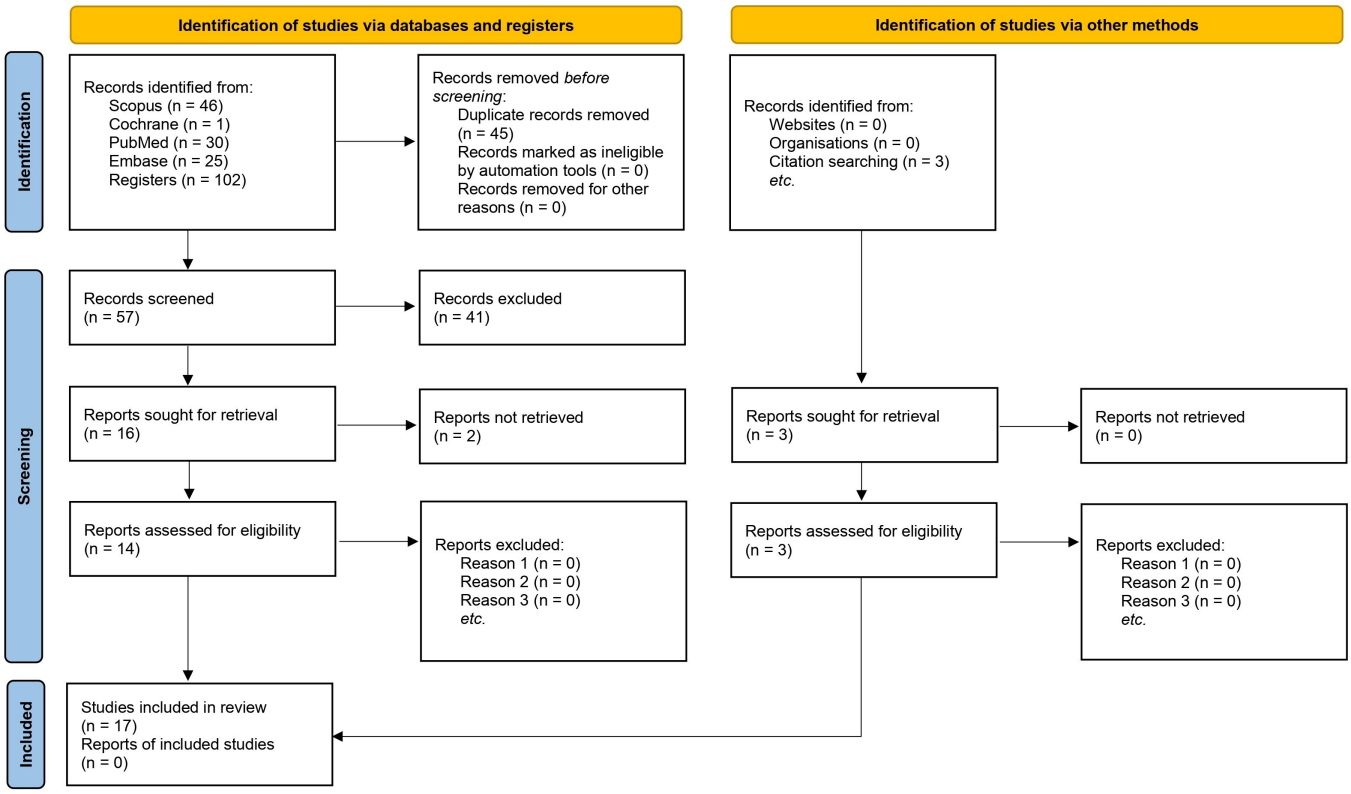

FIGURE 4. Flowchart based on PRISMA guidelines.

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.

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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.

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