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## Cowper's syringocele (bulbourethral cyst)

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Bulbourethral cyst or Cowper's syringocele (from the Greek "syringe" – tube, "cele" – expansion) is a cystic expansion of the excretory ducts of the bulbourethral glands (Cowper's glands). This disease is extremely rare and is more often diagnosed in the child population. The clinical manifestations of syringocele are non-specific and depend on many factors: size, localization, communication with the urethra, and the presence of an infectious component. Detection of syringocele is impossible without the use of special radiation and endoscopic diagnostic methods. Most often, this pathology is "masked" under inflammatory diseases of the genitals due to the fact that the symptoms are limited to secretions from the external opening of the urethra, changes in the general analysis of urine and dysuric manifestations. Currently in the domestic and foreign literature there are not even 20 publications devoted to this pathology.

**Keywords:** bulbourethral cyst; bulbourethral glands; Cowper's glands; syringocele.

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## Сирингоцеле Купера (бульбоуретральная киста)

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Сирингоцеле Купера (от греч. *syringo* — труба, *cele* — расширение), или бульбоуретральная киста, — это кистозное расширение выводных протоков бульбоуретральных желез (куперовых желез). Встречается данное заболевание крайне редко и чаще диагностируется у детей. Клинические проявления сирингоцеле Купера неспецифичны и зависят от многих факторов: размеров, локализации, сообщения с уретрой, наличия инфекционного процесса. Выявление сирингоцеле невозможно без применения специальных лучевых и эндоскопических методов диагностики. Чаще всего данная патология «маскируется» под воспалительные заболевания половых органов ввиду того, что симптоматика ограничивается выделениями из наружного отверстия уретры, изменениями в общем анализе мочи и дизурическими проявлениями. В настоящее время в отечественной и зарубежной литературе не наберется и 20 публикаций, посвященных данной патологии.

**Ключевые слова:** бульбоуретральная киста; бульбоуретральная железа; куперова железа; сирингоцеле.

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## INTRODUCTION

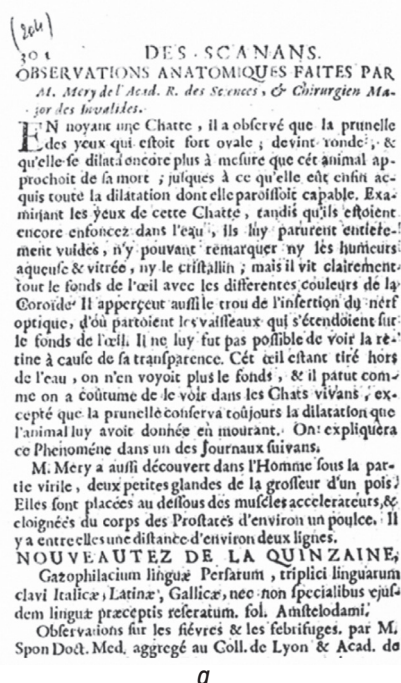
The bulbourethral glands were first mentioned by Jean Mery in 1684 (Fig. 1, *a*). They were further described in more detail by William Cowper (Fig. 1, *b*) in his work, "An account of two glands and their excretory ducts lately discovered in human bodies," published in the Proceedings of the Royal Society of London in 1699. Cowper depicted (Fig. 2) and described them as follows: "a quarter of an inch below the prostate gland, I found two other small glands located on both the sides of the urethra slightly above the bulb of the cavernous body. These glands have an oval shape, not exceeding the size of a small French bean" [1].

The bulbourethral glands are a pair of round organs, up to several millimeters in size, located posterior to the membranous urethra, approximately at the 3° and 9 o'clock positions, with excretory ducts opening in the bulbous urethra. Their main function is the release of pre-ejaculate into the male genital tract [2, 3].

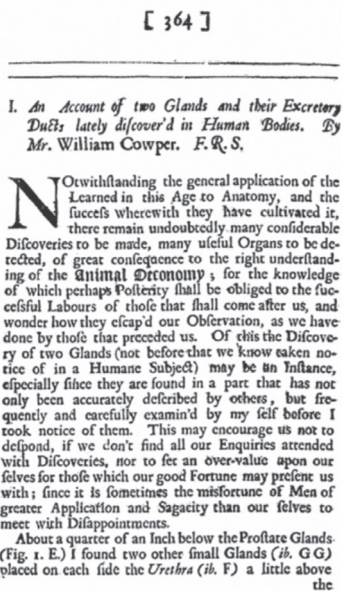
Cowper's syringocele was first described in 1983 by Maizels, who subsequently proposed to subdivide them into four types, namely, simple, perforated, non-perforated, and ruptured [4]. Among the four types, the

nonperforated one is the most difficult to recognize, which can be clinically asymptomatic in most cases. The simple, perforated, and ruptured types are differentiated only by the degree of dilatation of the outlet duct of the bulbourethral gland and the presence or absence of a "window" that connects it with the urethra. Bevers et al. [5] proposed a simplified classification (Fig. 3) of Cowper's syringocele, i.e., open and closed. The open type is characterized by pain in the perineum, discharge from the external urethral meatus, recurrent urinary tract infection, and post-micturition leakage. In the closed type, the most common signs include perineal pain, dysuria, and obstructive symptoms [6].

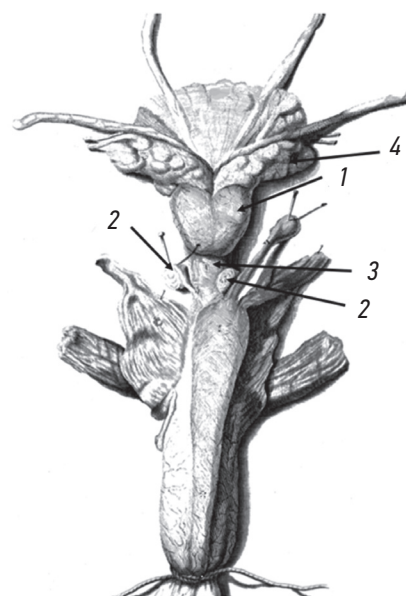
Campobasso et al. [7] analyzed the data of 15 patients with Cowper's syringocele and proposed to subdivide bulbourethral cysts into obstructive and nonobstructive depending on the clinical manifestations. The following pattern was revealed. For nonobstructive syringocele, the most typical symptoms include recurrent lower urinary tract infection, hematuria, fever, and post-micturition urinary leakage, while obstructive syringocele is characterized by signs of infravesical obstruction based on uroflowmetry and ultrasound examination.



a



b

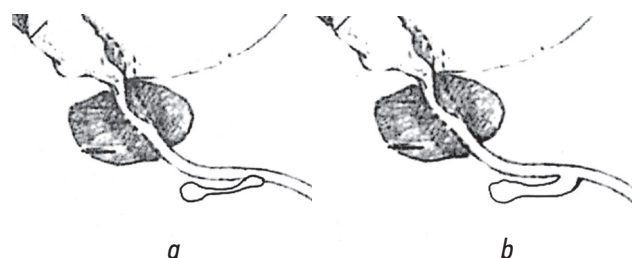


**Fig. 1.** Page of editions describing the bulbourethral glands: *a* – the first description of Jean Mery from the French edition of the Journal des scavans (1684); *b* – title page of W. Cowper's work "An Account of Two Glands and Their Excretory Ducts Lately Discover'd in Human Bodies" (1699)

**Рис. 1.** Страницы изданий с описанием бульбоуретральных желез: *a* – первое описание Jean Mery из французского издания «Journal des scavans» (1684); *b* – титульный лист труда W. Cowper «An Account of Two Glands and Their Excretory Ducts Lately Discover'd in Human Bodies» (1699)

**Fig. 2.** Organocomplex of the small pelvis of a man according (to: W. Cowper, 1699). 1 – prostate gland, 2 – bulbourethral glands, 3 – urethra, 4 – seminal vesicles

**Рис. 2.** Органокомплекс малого таза мужчины (по: W. Cowper, 1699). 1 – предстательная железа, 2 – бульбоуретральные железы, 3 – уретра, 4 – семенные пузырьки



**Fig. 3.** Types of Cowper's syringocele according to R. Bevers [5]: *a* – closed; *b* – open

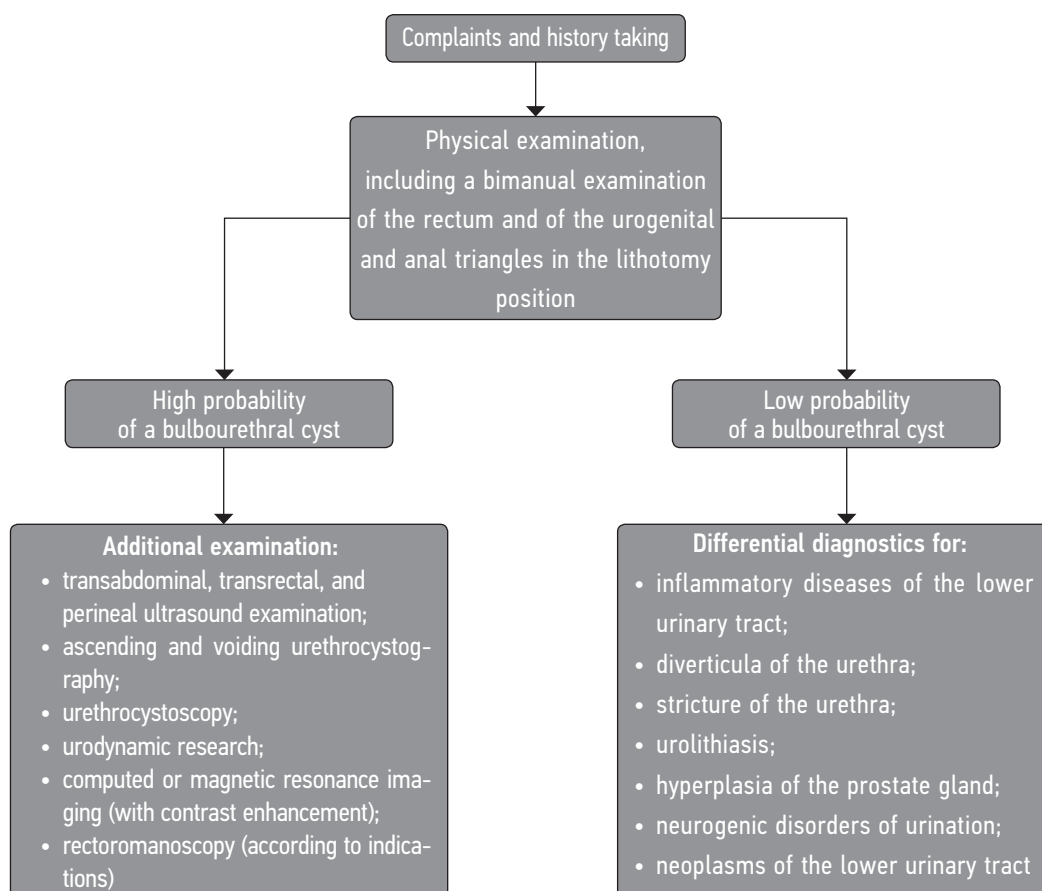
**Рис. 3.** Типы синрингоцеле Купера по R. Bevers [5]: *a* – закрытое; *b* – открытое

The clinical manifestations widely vary and depend mainly on its type. In patients with Cowper's syringocele, symptoms are similar to those of the lower urinary tract, such as urgency, post-micturition urinary leakage, urinary incontinence, recurrent urinary tract infection, hematuria, and urethral discharge. Post-micturition urinary leakage is more typical for the open type of syringocele, and its intensity depends on the configuration of the cyst, course, and depth of localization [5].

Diagnosis of a bulbourethral cyst is based on a thorough history taking and assessment of patient complaints, taking into account his age, supported by data

from radiation and endoscopic examination methods. Melquist et al. [6] proposed a simple algorithm for diagnosing syringocele. However, we recommend supplementing this approach with differential diagnostics with diseases such as urolithiasis, inflammatory diseases of the lower urinary tract and male genital organs, and urethral stricture, as well as with neoplasms of the pelvic organs (Fig. 4).

In patients with Cowper's syringocele, leukocyturia and bacteriuria are often detected in the general urine analysis and bacteriological culture of urine for microflora [8]. Ultrasound examination is informative for closed bulbourethral cysts if the cavity is determined.



**Fig. 4.** Proposed algorithm for the diagnosis of Cowper's syringocele

**Рис. 4.** Предлагаемый алгоритм диагностики синрингоцеле Купера

If the syringocele is open and emptied on examination, this diagnostic method may be not informative [9]. Magnetic resonance imaging (MRI) of the small pelvis, supplemented by urethral study, has high sensitivity and specificity, since it can visualize accurately the cyst cavity and determine its relationship with the urethra [10].

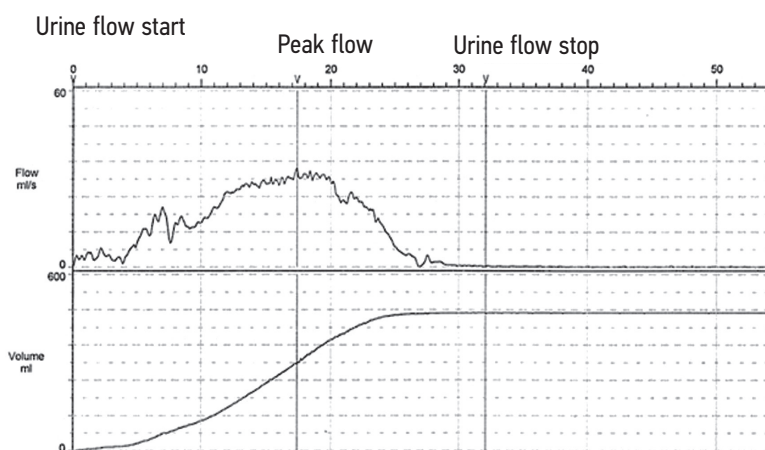
During urethroscopy, an open syringocele can be diagnosed when a “window” is found, which is a defect in the urethral wall. A closed syringocele is characterized by the prolapse of the cyst wall into the urethral lumen if the space-occupying lesion is filled sufficiently with liquid. Most authors tend to believe that ureteroscopy with MRI is the most informative method for diagnosing Cowper’s syringocele [6, 10]

A study revealed that asymptomatic bulbourethral cysts most often regress spontaneously and decrease in size, emptying in the course of conservative therapy. Moreover, symptomatic cysts require surgical intervention [11]. For the surgical treatment of Cowper’s syringocele, both open perineal approach and endoscopic techniques using electric and laser surgery are currently used, which include excision of the anterior wall of the bulbourethral cyst or its opening and marsupialization [12, 13]. Bevers et al. [5] reported about several patients who underwent transurethral dissection of the anterior wall of the bulbourethral cyst with a relapse-free follow-up period of 23 months. When the disease recurs after transurethral dissection of the anterior cyst wall, Santin et al. [14] suggested transperineal ligation of the ducts of the bulbourethral glands. Perineal excision of the bulbourethral cyst is reasonable only in large and recurrent cases after transurethral resection. Moreover, the authors did not indicate how large the cyst volume should be [15].

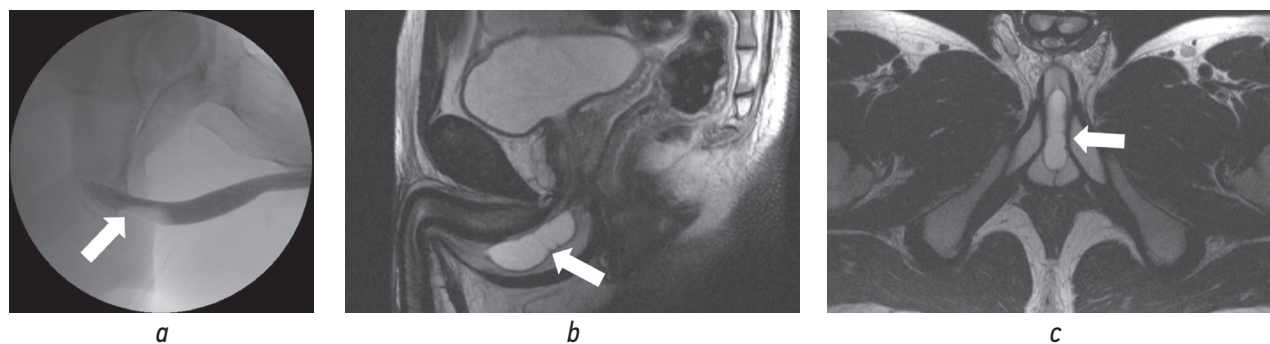
Very few studies have reported successful laparoscopic treatment of patients with Cowper’s syringocele and sclerotherapy of a closed-type cyst by the perineal method under ultrasound guidance and administration of a tetracycline group semi-synthetic antibiotic (minocycline hydrochloride) into its cavity. For laparoscopic excision of the bulbourethral cyst, Cerqueira et al. described their clinical experience of the surgical treatment of a large Cowper’s syringocele (10 × 10 × 8 cm; volume, 415 ml), prolapsing from the pelvis into the abdominal cavity and thus allowing the transperitoneal approach [16, 17].

## CLINICAL CASE

Patient Sh. (19 years old) was admitted to the urology clinic of the S.M. Kirov Military Medical Academy in August 2020 with complaints of intermittent, dull, and nagging pain along the urethra during urination and discomfort in the perineum. History assessment revealed that the above sensations occurred about 2 weeks ago after he experienced hypothermia. Digital rectal examination revealed the moderately enlarged prostate gland, without areas of fluctuation. Bimanual examination showed moderate tenderness in the perineum. Indicators of general clinical and biochemical blood tests were within the reference values. In the general analysis of urine, leukocyturia was determined, with up to 15–20 in the field of view. Bacteriological examination of urine showed the growth of *Corynebacterium glucuronolyticum* at  $5 \times 10^3$  CFU/ml. Ultrasound examination of the kidneys and urinary bladder did not detect pathological changes; the volume of the prostate gland was 23.1 cm<sup>3</sup>, and there was no residual urine. With uroflowmetry, the maximum urination rate was 33.7 ml/s, the average urination rate was 15.1 ml/s, the urination time was 32.3 s, and the volume of excreted urine was 468.7 ml (Fig. 5).

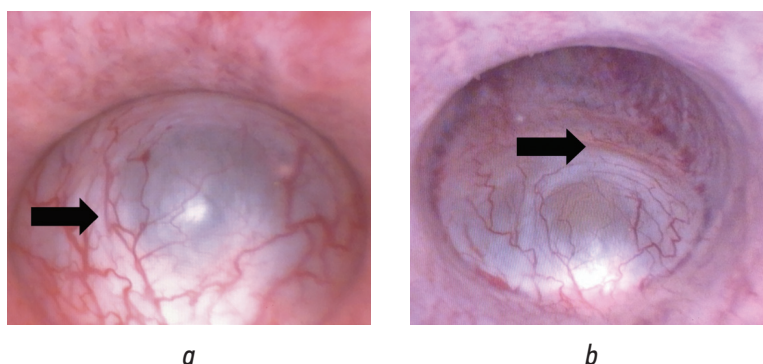


**Fig. 5.** Uroflowgram of patient Sh., 19 years old  
**Рис. 5.** Урофлоуграмма пациента Ш., 19 лет



**Fig. 6.** Patient Sh., 19 years old: *a* – ascending urethrograph of the filling defect corresponding to the location of the bulbourethral cyst is determined (indicated by the arrow); *b, c* – magnetic resonance imaging of the small pelvis, in two projections a cystic formation is determined – a syringocele with thin septa (indicated by arrows)

**Рис. 6.** Пациент Ш., 19 лет: *a* – восходящая уретрограмма, определяется дефект наполнения, соответствующий расположению бульбоуретральной кисты (указан стрелкой); *b, c* – магнитно-резонансная томограмма малого таза, в двух проекциях определяется кистозное образование – сириngoцеле с тонкими перегородками (указано стрелками)



**Fig. 7.** Urethroscopic picture of patient Sh., 19 years old: *a* – the anterior wall of the bulbourethral cyst; *b* – the septum in the cyst (indicated by arrows)

**Рис. 7.** Уретроскопическая картина пациента Ш., 19 лет: *a* – передняя стенка бульбоуретральной кисты; *b* – перегородка в кисте (указаны стрелками)

The patient was diagnosed with acute prostatitis. Antibacterial therapy (levofloxacin 500 mg once a day for 10 days), as well as anti-inflammatory therapy (diclofenac 100 mg per rectum for 7 days), and alpha-adrenoblocker therapy (tamsulosin 0.4 mg per day, 10 days) were started. The laboratory parameters were normalized; however, recurrent pain in the perineum during urination persisted.

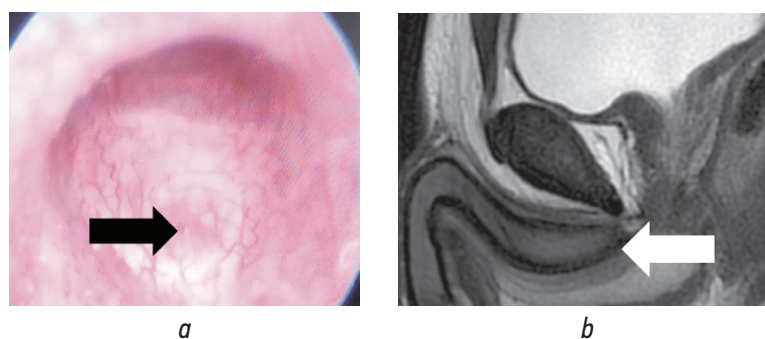
Given the persistent complaints, 2 months after discharge from the urological hospital, with arrested inflammation in the prostate gland, the patient was further examined for differential diagnostics according to the above algorithm with ascending urethrography, MRI of the pelvis and external genital organs, and urethroscopy (Fig. 6).

During ascending urethrography, the urethra was passable along the entire length; in the bulbous section along the ventral surface, a filling defect with clear even contours of up to 0.4 × 4.0 cm in size without leaks of contrast agent was determined, followed by the ingestion of the agent into the bladder (Fig. 6, *a*).

MRI revealed a cystic lesion located 3.5 cm distal to the bladder neck paraurethraly along the midline; it did not communicate with the urethra and measured 4.2 × 1.3 × 1.5 cm. The internal structure of the cyst was divided by three thin septa with a homogeneous fluid content. On ureteroscopy, in the bulbous section, the anterior wall of Cowper's syringocele prolapsed into the urethral lumen (Fig. 7).

Based on additional examination findings, the patient was diagnosed with a closed bulbourethral cyst. After conservative therapy with nonsteroidal anti-inflammatory drugs and alpha-adrenoblockers for 1 month, the symptoms regressed. After the third month of follow-up, urethroscopy showed a punctate "window" in the urethra, which was a sign of an open syringocele, and MRI showed the absence of previously registered liquid substance (Fig. 8)

Considering the absence of symptoms, presence of an open type of Cowper's syringocele, and young age, the patient was discharged under outpatient supervision.



**Fig. 8.** Patient Sh., 19 years old: *a* – urethroscopic picture of (the arrow indicates the “window” of the emptied cyst); *b* – control magnetic resonance imaging of the small pelvis without signs of cystic enlargement (the arrow indicates the place of the previously located cyst)

**Рис. 8.** Пациент Ш., 19 лет: *a* — уретроскопическая картина (стрелкой указано «окно» опорожненной кисты); *b* — контрольная магнитно-резонансная томограмма малого таза без признаков кистозного расширения (стрелкой указано место ранее расположенной кисты)

## CONCLUSION

Given the extremely rare incidence, there is currently no unified and clear algorithm for the diagnosis and treatment of patients with Cowper's syringocele. Notwithstanding, this pathology should be borne in mind when examining men, boys, and adolescents with signs of inflammatory diseases of the genitourinary system or symptoms of infravesical obstruction. For differential diagnostics of Cowper's syringocele, in addition to physical examination supplemented by bimanual examination, we recommend performing both radiation (ultrasonography and contrast-enhanced MRI) and endoscopic (urethroscopy) research methods following our modified examination algorithm.

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Conservative therapy can result in the regression of the cyst size or its spontaneous opening into the urethral lumen. When selecting a surgical treatment, the indications for surgery should be considered, which include severe pain syndrome, signs of infravesical obstruction, and cyst suppuration, proven by radiation diagnostics. In this case, endoscopic methods should be preferred as the first line of surgical treatment of Cowper's syringocele.

## ADDITIONAL INFORMATION

**Conflict of interest.** The authors declare no conflict of interest.

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