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## LETTER TO THE EDITOR

Male Fertility

# A case of obstructive azoospermia secondary to genitourinary tract infection caused by a prostatic utricle cyst

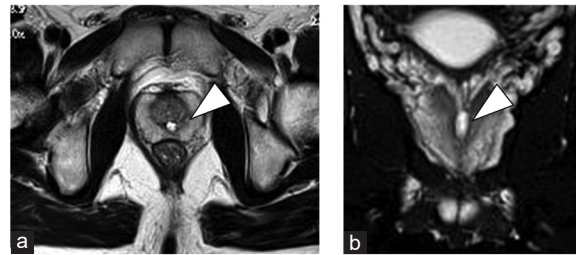
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Dear Editor,

Obstructive azoospermia (OA) is defined as the absence of spermatozoa in the sediment of a centrifuged sample of ejaculate and is usually caused by an obstruction anywhere along the posttesticular tracts. OA is diagnosed in 20%–40% of men with azoospermia. Although OA may be acquired congenitally, such as in Young's syndrome that presents with bilateral vas deferens abnormalities, it commonly develops as a sequela of prostatic and ejaculatory duct cysts, childhood inguinal hernias, epididymitis, and vasectomy. Acquired OA is rarely associated with multiple obstructions along both genital tracts. To the best of our knowledge, this is a rare case of OA to be reported with bilateral obstruction secondary to a genitourinary tract infection caused by a prostatic utricle cyst.<sup>1–3</sup>

This study was approved by the Institutional Ethics Committee of Yokohama City University Medical Center (Yokohama, Japan; approval No. B210300045). Informed consent form was obtained from the patient included in the study. A married couple (a 38-year-old man and a 30-year-old woman) consulted a gynecologist due to difficulty in conceiving for 10 months. The husband also complained of recurrent episodes of hematospermia. He denied any history of trauma or surgery. His semen analysis revealed azoospermia. He was subsequently referred to Yokohama City University Medical Center for further evaluation. Physical examination revealed no abnormalities in the penis, scrotum, testes, epididymides, or vasa deferentia. The patient's semen sample had a volume of 3.1 ml and did not contain spermatozoa. Serum testosterone, follicle-stimulating hormone, luteinizing hormone, and prolactin levels were 7.15 ng ml<sup>-1</sup>, 4.7 mIU ml<sup>-1</sup>, 5.0 IU l<sup>-1</sup>, and 15.2 ng ml<sup>-1</sup>, respectively. His karyotype was 46,XY. Microdeletion of the Y chromosome was not observed. Magnetic resonance imaging (MRI) and ultrasonography of the pelvis revealed a prostatic utricle cyst (**Figure 1**). MRI also revealed vasa deferentia and seminal vesicles bilaterally in the pelvis. OA was suspected, and the patient was willing to undergo reconstructive surgery if indicated. Therefore, intraoperative vasography was used to visualize



**Figure 1:** Prostatic utricle cyst in a 38-year-old patient with obstructive azoospermia: (a) axial view and (b) coronal view. The arrowheads indicate a prostatic utricle cyst.

the obstruction sites and aid in surgical reconstruction. Testicular sperm extraction (TESE) was considered in case reconstruction was not to be indicated. Intraoperatively, multiple adhesions were noted bilaterally in the following areas: between visceral and parietal layers of the tunica vaginalis, and both epididymal ducts. Other findings included thickening of the tunica vaginalis, and the dilatation of both ducts in the epididymis head near the efferent ducts. Vasography revealed an obstruction in the right vas deferens in the inguinal region. Contrast material was seen travelling freely through the left vas deferens, seminal vesicle, and prostatic utricle cyst. Microsurgical reconstruction was contraindicated because of multiple obstructions in both epididymal ducts and the right vas deferens. Spermatozoa were retrieved through conventional TESE and subsequent conception was achieved through intracytoplasmic sperm injection.

A prostatic utricle cyst is an area of focal dilatation within the prostatic utricle, which presents as a midline cystic mass in the male pelvis. While differentiating it from a Müllerian duct cyst may be difficult or even impossible, a prostatic utricle cyst may be appropriately visualized through ultrasonography and MRI.<sup>2,3</sup> Prostatic utricle cysts are associated with a variety of genitourinary abnormalities, such as hypospadias, cryptorchidism, and unilateral renal agenesis. In this case, considering the presence of bilateral epididymal obstruction with inflammatory changes and partial obstruction of the right vas deferens in the inguinal region, congenital absence of the vas deferens, epididymal duct atresia, and idiopathic epididymal obstruction were unlikely to cause OA. Although he had never experienced any subjective

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symptoms suggestive of a sexually transmitted disease, epididymitis, or prostatitis, prostatic utricle cysts are known to cause epididymitis, which may be due to retrograde infection of the urine remaining in the cyst or compression or obstruction of the ejaculatory duct by the cyst.<sup>4</sup> Consequently, the patient was diagnosed with OA caused by an asymptomatic genitourinary tract infection. Our observations suggest that genitourinary tract infections like epididymitis, although asymptomatic, may be a risk factor for OA development. While the extent of the role of genitourinary tract infection in OA is unclear, patients with prostatic utricle cysts who are diagnosed with genitourinary tract infections must be prescribed with antibiotics to decrease the risk of progression into OA. Future research may involve investigating bacteria that are most likely to be associated with OA and proposing appropriate antibiotic regimens accordingly. Research into alternative treatment options like transurethral resection of the ejaculatory ducts, transurethral ablation with the holmium laser,<sup>5</sup> and laparoscopic cyst resection<sup>6</sup> may also be performed.

Prostatic utricle cysts may also cause hematospermia.<sup>7</sup> Patients with OA who complain of hematospermia may require ultrasound imaging or MRI to exclude anatomical abnormalities like prostatic utricle cysts.<sup>2,3</sup> Our study highlighted the importance of preoperative ultrasonography, as it could help in avoiding unnecessary surgery if epididymal duct dilatations are detected on preoperative imaging.<sup>2,3</sup> In cases where preoperative ultrasonography shows normal results, as in our patient, intraoperative vasography and gross observation can help identify obstruction sites and determine whether surgical reconstruction or TESE is required. If the fructose and neutral glucosidase levels in the seminal plasma are approved, they can also aid in the preoperative diagnosis of OA. Overall, the need for surgical reconstruction must be heavily considered in patients with OA because sperm retrieval by TESE can help achieve the medical goal of conception equally well. In this case, reconstruction was challenging due to long-term obstruction in the inguinal segment of the right vas deferens and additional obstructions in both epididymides. Epididymal vasovasostomy is also a surgical alternative but has low success rates. To the best of our knowledge, this is a rare case of acquired OA that appeared to be secondary to genitourinary tract infections caused by a prostatic utricle cyst.

## AUTHOR CONTRIBUTIONS

TS and MK contributed to the project development, data collection, and manuscript writing. KU, SK, TT, KT, MI, and YK contributed to the data collection. YY contributed to the project development. All authors read and approved the final manuscript.

## COMPETING INTERESTS

All authors declare no competing interests.

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