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Case Report

# Similar Mullerian Cysts in an Identical Twin: A Case Report and Literature Review

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## Abstract

Several genital anomalies have been reported in the identical twins which have a tremendous effect on reproductive status. No previous studies have reported the Mullerian duct cyst in identical twin brothers. We describe a rare case of Mullerian cyst in a male identical twin with infertility. A 43-year-old man presented with 2 years of infertility. In the spermogram analysis, sperm count leaded to azoospermia detection. Transrectal ultrasonography (TRUS) examination was done. An echo-free structure in the mid part of prostate suggested a Mullerian cyst which had caused ejaculatory duct obstruction. The other twin, who dealt with infertility as well, was referred for TRUS. A Mullerian cyst was detected. Ultimately, testicular sperm extraction and percutaneous epididymal sperm aspiration procedures were recommended. Imaging with variety ranges of modality can help to identify Mullerian cyst. Further researches for detecting the genetic factor causes of this anomaly should be considered.

**Keywords:** Imaging, identical twin, infertility, Mullerian cyst

## Introduction

Twin birth accounts for approximately 3% of all childbirths and 30% of twins are monozygotic.[1] Monozygotic or identical twin stems from the division of a unique egg after fertilization.[2] Several genital anomalies have been reported in the identical twins which have a tremendous effect on reproductive status. Mullerian duct-related abnormalities have been reported in female twins.[3] To the best of our knowledge, no previous studies have reported the Mullerian duct cyst in identical twin brothers. We aim at present a case of Mullerian duct and its other related complications in a male identical twin referred for infertility assessment.

## Case Report

A 43-year-old man presented with 2 years of infertility. The medical history was normal and he was the first birth of an identical twin. In spermogram analysis, sperm count leaded to azoospermia detection. Other hormonal profiles, namely follicle-stimulating hormone, luteinizing hormone, prolactin, and testosterone were in the normal ranges.

He was referred to the ultrasonography ward for further assessment and undergone Doppler examination of the scrotum. Two-dimensional color Doppler examination of the scrotum showed that both testes were normal homogeneous parenchyma. It revealed that the blood flow and size of the testes were normal and there were no significant pathology in the epididymis and the cord of epididymis.

Then, transrectal ultrasonography (TRUS) examination was done. The TRUS of prostate and seminal vesicles was done by Bi-plane transducer. The prostate measurement was 39 mm × 20 mm × 34 mm. The parenchymal echogenicity was heterogen. An echo-free structure with 8 mm × 10 mm diameter in the mid part of prostate suggested Mullerian cyst which had caused ejaculatory duct obstruction [Figure 1]. Both seminal vesicles were seen with heterogen echogenicity and cystic formation. Both right and left vas deferens were dilated.

The other twin, who dealt with infertility as well, was referred for TRUS. The prostate size was 39 mm × 25 × mm × 35 mm. The parenchymal echogenicity was nonhomogeneous and a Mullerian cyst with 13 mm × 15 mm diameters was

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detected [Figure 2]. The seminal vesicles were seen with nonhomogeneous echogenicity and cystic formation. Both right and left vas deferens were dilated as well.

Considering the fact that the urinary anomalies occur along with the genitalia anomalies, urinary assessment with abdominal ultrasound examination was carried out and no kidney and bladder anomaly were reported.

Ultimately, testicular sperm extraction and percutaneous epididymal sperm aspiration procedures were recommended. The inform consent forms were filled with the patients.

## Discussion

Mullerian cyst stems from the embryonic Mullerian duct. It affects both females and males, as well.[4] The Mullerian cyst is located in various sites. Consequently, no exact prevalence has been reported so far. However, it is estimated that <1% of women suffer from Mullerian cyst.[5]

Similarly, the locations, manifestations, and effects of Mullerian cyst would be different in men. The incidence of Mullerian cyst in male children is reported approximately 1% and the manifestations, mostly peak at 20–40 years old.[6]

The main complaint would be subfertility. However, other comorbidities and potential hazardous conditions should not be neglected.[7] A brief review of the main findings of recent Mullerian cyst case reports in men and their management are revealed in Table 1.

To the best of our knowledge, to date, no previous study has reported a twin brother with the same Mullerian duct cysts. However, a twin sister with the same genital anomalies, amenorrhea, and infertility complaint was reported (Sreedevi NS, 2008).

They mentioned that genetic factors result in such a rare anomaly in twins. Both of twin sisters had other comorbidities such as diabetes. They revealed that genetic factors and changes

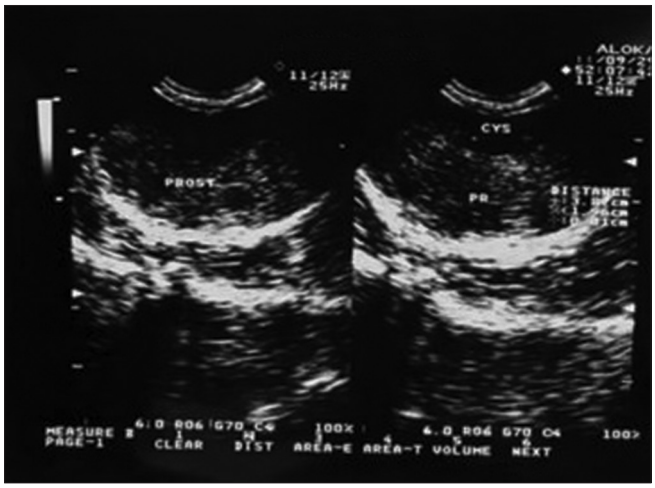


**Figure 1:** Mullerian cyst in first twin

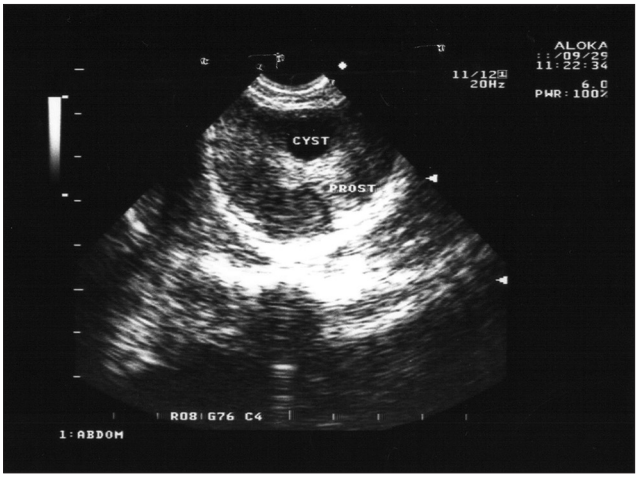


**Figure 2:** Mullerian cyst in second twin

**Table 1: Literature review of the cases of Mullerian duct cysts**

| Reference number | Year | Age | Site | Size (mm) | Treatment | Histology |
|---|---|---|---|---|---|---|
| [8] | 2006 | 9 months | Left side of the scrotum with acute epididymitis | 15×13 | Surgery | Benign unilocular cyst lined with columnar epithelium |
| [9] | 2008 | 39 | By the prostate | - | Robot-assisted removal | Mullerian cyst |
| [10] | 2009 | 77 | Pelvic | Large | Open surgical resection | Tratified cubocolumnar cells |
| [11] | 2017 | 39 | Behind the bladder | Large | Transurethral drainage | Mullerian cyst |
| [12] | 2017 | 15 | Posteriorly to the prostatic urethra | 7.5*8.5 | Conservative | - |
| [13] | 2018 | 43 | Posterior to the bladder and prostate | 80 | Template-guided transperineal aspiration | Mullerian duct cysts |
| [14] | 2019 | 23 | Retroperitoneum | 33 | Surgery and chemotherapy | Cytokeratins |
| [15] | 2019 | 20 | Posterior to the right kidney | 22828 | Surgical resection | Simple columnar cells with multiple intracytoplasmic vacuoles |
| [4] | 2021 | 17 | Scrotum | 47*17 | Surgical resection | Tall columnar |
| [16] | 2021 | 22 | Pelvis and recto vesical space | 125*75*65 | Laparoscopic right orchidectomy (streak) + Mullerian remnant excision + left orchidopexy + first-stage hypospadias repair | Known case of 45XO/46XY mixed gonadal dysgenesis, Mullerian remnants |

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in hormonal balance in developmental stages might contribute to developmental and congenital anomalies, especially in the gastrointestinal and urogenital tracts.[3]

The male twin is not excluded from these rules and if there is any basic genetic factor which causes the hormonal imbalance and developmental distortion, it will occur in the both of identical twin brothers.

Since the developmental pathways of the male and female genital organs are different during pregnancy, the Mullerian duct anomalies, their manifestations and later consequences vary.

Depending on the size of Mullerian cyst and whether an infection is present or not, the symptoms of Mullerian cyst in the male range from urinary disturbances to hematuria. If the cyst is large enough, obstructive effects and systemic effects such as abdominal pain, fever, and nausea are seen.[4] It might relate to malignancy, occasionally.[6]

When it comes to long-term effects, the subfertility is propounded inasmuch the compressing effect of a cyst results in ejaculatory duct obstruction.[17] In such cases, the hormonal profiles are normal; however, the obstruction contributes to azoospermia or oligospermia.[7]

The ultrasonography is said to be in the vanguard for detecting the Mullerian cysts.[18] The main features which draw attention to the Mullerian cyst in the ultrasonography are a nonhomogenous and echo-free structure in the prostate, ejaculatory duct obstruction, seminal vesicle cyst, and vas deference dilation. The congenital prostate cyst has a regular shape with distinct borders. In cases that multiple cysts are seen and the edges are not regular, hyperplasia of prostate is propounded.[4]

TRUS is said to be most useful modality for detection of exact congenital anomalies in infertile men, especially in azoospermia or hematospermia. TRUS is a safe and noninvasive method, so it is highly recommended for assessing the lesions of the prostate, seminal vesicles, and the ejaculatory ducts.[19-21]

If the ultrasonography is not sufficient for definite detection of Mullerian cyst, computed tomography is used to depict the exact size, location, and position of the cyst. Otherwise, magnetic resonance imaging (MRI) might be utilized for ruling out the different probable anomalies.[22] Using three-dimensional capability, MRI is able to depict the exact tissues and their contrasts.[4]

## Conclusion

The Mullerian cyst is considered as a rare cause of male infertility. Provided that it is detected timely, the infertility can be treated with advanced surgical methods and assisted reproductive technology. Imaging with variety ranges of modality can help to identify Mullerian cyst. Further researches for detecting the genetic factor causes of this anomaly should be considered.

### Declaration of patient consent

The inform consent forms were filled by twin brothers. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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### Conflicts of interest

There are no conflicts of interest.

## References

1. Chasen ST, Chervenak FA. Twin pregnancy: Prenatal Issues. Waltham, MA: UpToDate; 2017. Available from: http://enjoypregnancyclub.com/wp-content/uploads/2017/05/Twin%20pregnancy%20Prenatal%20issues.pdf. [Last accessed on 2016 Aug 18].
2. Jain AK, Prabhakar S, Pankanti S. On the similarity of identical twin fingerprints. Pattern Recognit 2002;35:2653-63.
3. Sreedevi NS KM, Vinayachandran S, Chandrika CV, Krishnan Sandhya MP. Twin sisters with primary amenorrhea. J Obstet Gynecol India. 2008;58:3.
4. He J, Tang K. A Müllerian cyst in a male adolescent: A case report and literature review. J Int Med Res 2021;49:3000605211016663.
5. Töz E, Sancı M, Cumurcu S, Özcan A. Müllerian cyst of the vagina masquerading as a cystocele. Case Rep Obstet Gynecol 2015;2015:376834.
6. Suzuki S, Furukawa S, Kyozuka H, Watanabe T, Takahashi H, Fujimori K. Two cases of paraovarian tumor of borderline malignancy. J Obstet Gynaecol Res 2013;39:437-41.
7. Hendry WF, Pryor JP. Müllerian duct (prostatic utricle) cyst: Diagnosis and treatment in subfertile males. Br J Urol 1992;69:79-82.
8. Donkol RH, Monib S, Moghazy K. Müllerian duct cyst as a cause of acute infantile-onset epididymitis. Pediatr Radiol 2006;36:1197-9.
9. Selli C, Cavalleri S, De Maria M, Iafrate M, Giannarini G. Robot-assisted removal of a large seminal vesicle cyst with ipsilateral renal agenesis associated with an ectopic ureter and a Müllerian cyst of the vas deferens. Urology 2008;71:1226.e5-7.
10. Jaidane M, Hidoussi A, Slama A, Hmida W, Sorba NB, Mosbah F. An acute urinary retention in an old man caused by a giant Müllerian duct cyst: A case report. Cases J 2009;2:203.
11. Shabaninia S, Yahyazadeh SR. Symptomatic müllerian duct cyst in a male adult: A rare case report. Acad J Surg 2017;4:24-6.
12. Feutry G, De Perrot T, Wirth GJ, Montet X, Martin SP. Prostatic utricle cyst as the most likely cause in a case of recurrent episodes of hematospermia. Case Rep Urol 2017;2017:7502878.
13. Yogeswaran C, Srinivasan V, Ekwueme KC. Mullerian duct cyst treated with template-guided transperineal aspiration: A case report and review of the literature. JRSM Open 2018;9:2054270417725497.
14. Naem A, Dlewati A, Alhimyar M, Ousta MA, Alsaid B. A rare presentation and recurrence of a retroperitoneal Müllerian cyst in a male patient: A case report. Int J Surg Case Rep 2019;65:301-4.
15. Tolani MA, Bello A, Ahmed M, Maitama HY. Symptomatic Müllerian cyst in the retroperitoneum of an adult male: Case report and review of the literature. Archives of Medicine and Surgery. 2019;4:53.
16. Ravirajendran S, Palaniyandi V, Arora A, Ramanan V, Kumaresan N. Mullerian remnants presenting as a pelvic cyst in a young adult with 45X0/46XY mixed gonadal dysgenesis. Urol Ann 2021;13:76-9.
17. Dimasis N, Koukourikis P, Klampatsas A, Xirou P, Sountoulides P. A unique case of aggressive uterine cancer in a 45-year-old man with persistent Müllerian duct syndrome. Arch Esp Urol 2019;72:435-8.
18. Bohîlţea RE, Cîrstoiu MM, Turcan N, Ionescu CA. Ultrasound diagnostic of mesonephric paraovarian cyst – Case report. J Med Life 2016;9:280-3.
19. Mayersak J, Kuchenbecker DA. Müllerian duct cyst: Case report with diagnosis established by transrectal prostatic ultrasound. Urology 1993;41:176-80.
20. Razek AA, Elhanbly S, Eldeak A. Transrectal ultrasound in patients with hematospermia. J Ultrasound 2010;13:28-33.
21. Jarow JP. Transrectal ultrasonography of infertile men. Fertil Steril 1993;60:1035-9.
22. Kawaguchi M, Kato H, Hara A, Suzui N, Tomita H, Miyazaki T, *et al.* CT and MRI characteristics for differentiating mediastinal Müllerian cysts from bronchogenic cysts. Clin Radiol 2019;74:976.e19-25.