Cystic dysplasia of the rete testis in a young adult accompanying ipsilateral renal agenesis: A case report

Ivan Franin (franin.ivan@gmail.com)
Sestre milosrdnice University Hospital Centre

Igor Grubišić
Sestre milosrdnice University Hospital Centre

Lana Postružin Gršić
Sestre milosrdnice University Hospital Centre

Monica Stephany Kirigin
Sestre milosrdnice University Hospital Centre

Tonči Vodopić
Varaždin General Hospital

Božo Krušlin
Sestre milosrdnice University Hospital Centre

Case Report

Keywords: rete testis cystic dysplasia, renal agenesis, immunohistochemistry

Posted Date: December 29th, 2022

DOI: https://doi.org/10.21203/rs.3.rs-2397397/v1

License: This work is licensed under a Creative Commons Attribution 4.0 International License. Read Full License
Abstract

**Background:** A 31-year-old man was referred from an outside institution to the department of urology under the suspicion of a testicular tumor, with left-sided testicular pain lasting a couple of months in duration. Physical examination showed a hard, thickened and small left testis on palpation with diffuse, inhomogeneous ultrasonographic appearance. After urologic examination a left-sided inguinal orchiectomy was performed. The testis, epididymis and spermatic cord were sent to pathology.

**Case presentation:** On gross examination, a cystic cavity filled with brown fluid content and surrounding brownish parenchyma measuring up to 3.5 cm in diameter was found. Histologic examination showed cystically dilated rete testis lined with cuboidal epithelium and a positive immunohistochemical reaction to cytokeratins. The cystic cavity was microscopically a pseudocyst filled with extravasated erythrocytes and abundant clusters of siderophages. The siderophages extended into the testicular parenchyma, surrounding the seminiferous tubules and spreading out around ducts of the epididymis, which were also cystically dilated with siderophages inside their lumina.

**Conclusions:** On the basis of clinical data, histological and immunohistochemical analysis, a diagnosis of cystic dysplasia of the rete testis was established. According to the literature there is a very well-known association between cystic dysplasia of rete testis and ipsilateral genitourinary anomalies. The patient was referred to the department of radiology, and a multi-slice computed tomography scan revealed ipsilateral renal agenesis, right seminal vesicle cyst reaching up to the iliac arteries and a multi-cystic formation cranial to the prostate.

**Background**

A rare, benign cause of testicular mass in children is cystic dysplasia of the rete testis. Around 60 cases have been recorded since Leissring and Oppenheimer initially described it in 1973. [1] Cystic dysplasia of the rete testis is characterized by irregular cystic areas in the mediastinum or rete testes that are lined by cuboidal epithelium. It frequently coexists with genitourinary tract abnormalities, most prominently renal agenesis. [2]

**Case Presentation**

A 31-year-old male patient was referred to the department of urology, who had been experiencing left-sided testicular pain for a few months and was suspected of having a testicular tumor. On palpation the left testis was firm, thickened and small. A widespread, inhomogeneous ultrasonographic appearance was seen in the left testicle. A left sided inguinal orchiectomy was performed. The spermatic cord, epididymis and testis were sent to pathology. Upon gross inspection, a 3.5 cm-diameter cystic cavity with brown fluid content and surrounding brownish parenchyma was discovered in the left testis (Fig. 1). Histologic examination of the tissue revealed cystically dilated rete testis lined with cuboidal epithelium, which showed a positive immunohistochemical reaction to cytokeratins. Microscopically, the cystic cavity...
contained extravasated erythrocytes and large clusters of siderophages. The epididymis' ducts were also
cystically dilated with siderophages inside their lumina, and the testicular parenchyma's seminiferous
tubules were also surrounded by siderophages (Fig. 2a,b). The diagnosis of cystic dysplasia of the rete
testis was made on the basis of clinical information, histological examination and immunohistochemical
analysis. The relationship between ipsilateral genitourinary abnormalities and cystic dysplasia of the rete
testis is well-known in the literature. The patient was referred to the radiology department, where a multi-
slice computed tomography scan of the patient revealed ipsilateral renal agenesis, a right seminal vesicle
cyst reaching up to the iliac arteries, and a multi-cystic formation cranial to the prostate (Fig. 3).

Discussion And Conclusions

The precise etiology of cystic dysplasia of the rete testis is still unknown. According to Leissring and
Oppenheimer, the mediastinum testis gradually degenerates into small cysts as a result of the lack of a
connection between the mesonephric duct and germinal epithelium at the level of the rete testis. The
associated urinary tract problems may also be explained by this theory. The ureteral bud, from which the
mesonephric duct develops, eventually gives rise to the kidney. Another theory suggested by Nistal et al.
involved the excessive fluid secretion in immature seminiferous tubules without a lumen. [3] The gradual
canalization of the tubules during childhood may be the cause of the cysts' spontaneous regression. [4]

The age at presentation of cystic dysplasia of rete testis ranges from birth to 18 years, with a median of
5.2 years. Only two cases found in the literature included adults, aged 23 and 63 years, but were excluded
in a review study by Contini et al, the reason being that they were adults. [2] Our patient would therefore
be the second oldest patient by age at presentation of cystic dysplasia of the rete testis reported in the
English literature. Our patient presented with testicular pain, which was present in 9% of patients reported
until this day. The most common associated urogenital system anomaly with cystic dysplasia of rete
testis is ipsilateral renal agenesis (50%), which was also observed in our patient. Alongside the ipsilateral
renal agenesis we observed enlarged seminal vesicles, which were present in 4,5% of other cases. 48.5%
of the other patients underwent orchiectomy as well as our patient. [2] Surgery should only be used in
cases where the diagnosis is ambiguous, according to a diagnostic and therapeutic protocol proposed by
Helman et al. [5] For patients who fit the criteria for cystic dysplasia of the rete testis yearly scrotal
ultrasonography was suggested. [5] Close monitoring during the initial several months following
diagnosis is essential when non-surgical therapy is chosen. However, a definite diagnosis and the
exclusion of malignant cystic testicular lesion require surgical biopsy and histological confirmation,
particularly when the lesion does not regress on ultrasound. [2]

To conclude we suggest having cystic dysplasia of the rete testis in mind as a possible differential
diagnosis in adults because of the late presentation of this lesion in our patient.

Declarations
Ethics approval and consent to participate: This case report is in compliance with all ethical standards and is upholding integrity of the journal's scientific record.

Consent for publication: Consent for publication was obtained.

Availability of data and materials: All data and materials, including pathology slides and tissue blocks can be found at our Department.

Competing interests: The authors have no relevant financial or non-financial interests to disclose.

Funding: None.

Contributions: Ivan Franin (searching for references, writing the report, material preparation, data collection and analysis), Igor Grubišić (providing clinical information of the patient), Lana Postružin Gršić (providing radiology images of the patient), Monica Stephany Kirigin (proofreading, lecturing), Tonći Vodopić (using digital pathology software for taking microscopic images), Božo Krušlin (interpretation of H&E and immunohistochemistry slides, reaching the diagnosis, mentoring). All authors contributed to the study conception and design, all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

Acknowledgements: None.

Authors' information: None.

Footnotes: None.

References


Figures
Figure 1

Gross examination showing brownish testicular parenchyma and a cystic cavity filled with fluid.
Figure 2

a H&E stain showing dilated rete testis. b Cytokeratins stain showing dilated rete testis.
Figure 3

Multi-slice computed tomography using gastrografin showing renal agenesis and (multi)-cystically dilated seminal vesicle.