CONGENITAL PARAMEATAL URETHRAL CYST IN THE MALE: A CASE REPORT AND REVIEW OF LITERATURE

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Case Report:

CONGENITAL PARAMEATAL URETHRAL CYST IN THE MALE: A CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Parameatal urethral cyst is a scarce congenital condition that was first reported in two males in 1956, until now in reported literature only found less than 50 cases, in both adults and children. Our patient, case of parameatal urethral cyst in a 5 years old boy is reported. Complete excision with total removal of the epithelium of the cyst is required management for the treatment and prevention of cyst reocurrance. A 5 years old male with a cystic lesion around urethral meatus since birth. At least 5 month the parents complain distorted urinary flow and poor appearance, and no other urinary symptom, no history of trauma. On physical examination, cystic mass with spherical shape which was about 0.5 cm in diameter was found around external meatus. There was no inflammatory sign. And there was normal blood laboratory (blood counts and blood chemistry) and urine laboratory (urine analysis and urine culture). The patient undergo completely excision of the cyst under general anaesthesia, and remove all of the lining epithelium. Good appearance results were obtained after 2 months follow up, without meatal strictures and urine stream problems, and no postoperative complications or recurrence. Pathological : Squamous epithelial, granulation tissue with chronic inflamation. Parameatal urethral cyst is a very rare benign condition that is asymptomatic in most of the cases. It may be present since birth or appear later and is prevalent in young males. Its etiology remains unclear and treatment is by complete surgical excision to avoid complications and recurrence. Good cosmetic results were obtained in this case without any recurrence at two months follow up.

***Keywords**: Parameatal cyst; glans penis; external meatus urethra; urethral cyst*

ABSTRAK

Kista parameatal uretra adalah kelainan kongenital yang sangat jarang. Kasus pertama dilaporkan pada dua laki-lahi tahun 1956 oleh Thompson dan lantin, tidak lebih dari 50 kasus telah dilaporkan, baik pada dewasa maupun anak-anak. Kita melaporkan satu kasus kista parameatal uretra pada anak laki-laki usia 5 tahun. Eksisi dilakukan dengan membuang keseluruhan kista bersama epitelnya untuk mencegah terjadinya kekambuhan. Anak laki-laki berusia 5 tahun dengan kista pada meatus uretra sejak lahir. Orang tua pasien mengeluhkan kencing bercabang dan kosmetik yang jelek sejak 5 bulan terakhir. Selain itu tidak ada keluhan kencing, riwayat trauma pula disangkal. Hasil dari pemeriksaan fisik didapatkan massa kista bulat pada meatus uretra eksterna dengan diameter 0,5 cm. tidak didapatkan tanda-tanda inflamasi. Pemeriksaan laborat seperti darah, analisa kencing dan kultur urin normal. Eksisi kista telah dilakukan dibawah anestesi umum, dengan mengeksisi epitel disekitarnya. Kosmetik yang bagus diamati dalam 2 bulan follow up tanpa striktur meatus uretra dan gangguan aliran kencing. Dari patologi didapatkan squamous cell, jaringan granulasi dengan kronik inflamasi. Kista parameatal uretra adalah kasus yang jarang ditemukan, dan biasanya asimtomatik. Ini dapat muncul sejak lahir, atau muncul beberapa saat dan prevalensinya pada laki-laki muda. Etiologi dan penyebab terjadinya masi belum jelas dan pengobatan pada penyakit ini adalah eksisi kista untuk menghindari komplikasi dan kekambuhan. Hasil konsmetik yang bagus dilaporkan diamati pada kasus ini tanpa kekambuhan pada 2 bulan follow-up.

***Kata kunci**: Kista parameatal; gland penis; meatus uretra eksterna; kista uretra*

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INTRODUCTION

Parameatal urethral cyst is an abnormal disorder which usually seen at birth, after circumcision, or in the second decade of life. These cysts have round shape and are adjacent to the urethral meatus and can cause obstruction in urethral tract (Kennedy 2010). These cysts are rarely reported, less than 50 cases reported. The first case reported by Thompson and Lantin in 1956 (Nerli et al 2012, Mahato et al 2014) Parameatal urethral cysts are often asymptomatic, but some case can cause problem in urinary system, dysuria, branched urine, urinary retention, and even pain during sexual intercourse and poor cosmetics (Mahato et al 2011). Until today, the etiology of parameatal urethral cysts is not known. Shiraki suggests that these cysts may be the result of occlusion of the paraurethral ducts or there are several studies suggesting the failure in separation of preputium from the glans along the coronary sulcus can cause this disease (Wein et al, 2012). In this paper, we will present a case of parameatal urethral cyst in a 5 years old boy. This case report has received a certificate of ethical clearance from Komite Etik Penelitian Kesehatan RSUD Dr. Soetomo with Ref. No: 0114/LOE/301.4.2/IX/2020.

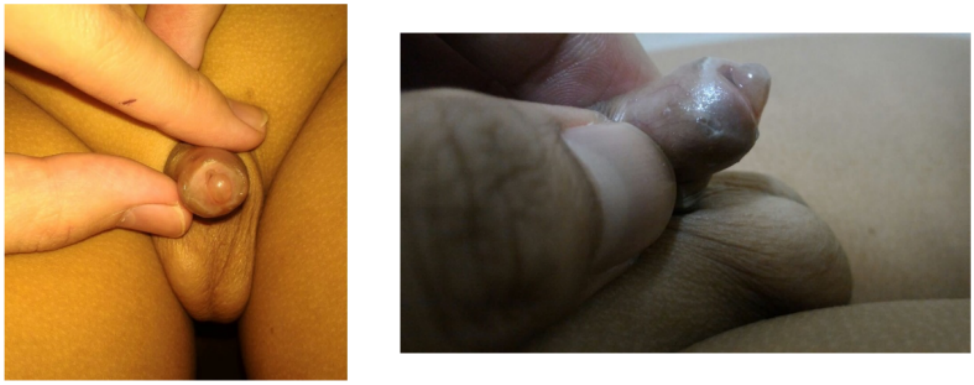
CASE REPORT

A 5-year-old boy brought by his parents to Urology Clinic at RSAL Dr. Ramelan Surabaya, with complaints of branching urine since 1 week ago. Initially, there is a lump that filled like water appeared on the tip of the penis since birth, which grew bigger with age. Other symptoms such as pain when urinating and fever are not felt by the patient. There is no history of trauma and history of infection.

On physical examination, there was a cystic mass in the external urethral meatus with a diameter of 0.5 cm, no signs of inflammation were seen in Figures 1a and 1b. From laboratory tests such as complete blood count, kidney function (RFT), urinalysis, and urine culture, there were also no abnormalities.

In this case, excision of the cyst and circumcision was performed using general anesthesia. The patient was observed for 2 months, and there was no recurrence, no problem when patient urinate and the flow of urine is normal (Fig. 2).

From the histopathology, it was found that the tissue was covered by squamous epithelium, in the stroma there was a proliferation of small to moderate blood vessels covered with a layer of endothelial containing erythrocytes in the lumen. There were a lot of PMN inflammatory cells and mononuclear infiltration around the tissue. With the conclusion of granulation tissue with chronic inflammation.



1a **1b**

Fig. 1a, b. Parameatal cyst of urethra in 5-year-old boy.

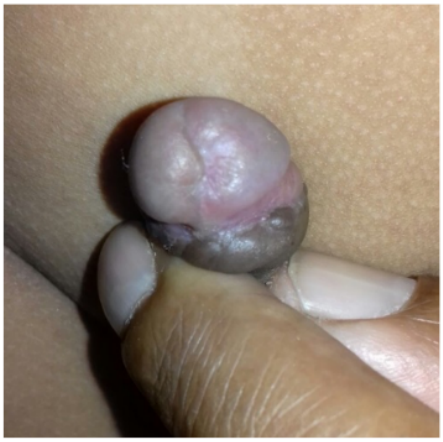


Fig. 2. Evaluation 2 weeks after surgery, complaint (-), urine smoothly and strong urine stream.

DISCUSSION

Parameatal urethral cyst is a rare case, it can be found in both men and women. In women these cases were reported to be 1 in 7246 births (Herek et al 2000, Phupong & Aribarg 2000). These cysts appear spontaneously at the onset of birth, or the second decade of life, on prepubertal in males, while in females often appeared at neonatal age. The etiology of parameatal cysts is still not fully understood, obstruction of the paraurethral duct, either spontaneous or secondary to infection is thought to be one of the factors that cause parameatal cysts. Some authors believe that parameatal urethral cysts occur in interfering process with the division of foreskin from the glands of the penis and some believe that cyst formation results from anomalous fusion of the urethra, formation of median raphe cysts, or caused by obstruction of the paraurethral duct (Gupta & Gupta 2015, Lal & Agarwal 2013, Willis et al 2011, Ichiro et al 1985).

Some cysts are associated with inflammation leading to infection and trauma leading to obstruction of the parameatal duct and causing parameatal cysts. In the study conducted by Willis et al (2011), only 1 in 18 patients had an inflammatory process in their histopathology.

Ichiyanagi et al (2001) detected the presence of prostate specific antigen (PSA) on cells from parameatal urethral cysts, and the hypothesis was these cysts originated from accessory glands in the urethra. Soyer et al reported two female infants with parameatal urethral cysts, allegedly related to the role of estrogen in the development of the unborn baby (Lal & Agarwal 2013, Gupta & Gupta 2015).

Perimeatal cysts are usually less than 1 cm in size, and are located on one side at the urethral meatus, but bilateral cases have also been reported. Parameatal cysts often show no symptoms, but some of the symptoms that can appear are urinary disorders such as branching urine, pain when urinating, or even urinary retention, pain during sexual intercourse, and bad cosmetics are reasons to come to the doctor for the first time (Gupta & Gupta 2015, Lal & Agarwal 2013).

Conservative

Parameatal urethral cysts in neonates who are at high risk for surgical management are expected to resolve spontaneously (Fujimoto et al 2007). The timing of conservative management is not clearly defined in several studies. If cysts do not disappear within 6 months, surgical intervention is recommended. However, the recurrence rate from this conservative treatment is still high (Fujimoto et al 2007, Neeli et al 2012).

Needle aspiration

The disadvantages of needle aspiration are recurrence and the satisfaction in cosmetics is inconclusive (Fujimoto et al 2007, Neeli et al 2012).

Marsupialization

The marsupializtion has been reported to have unsatisfactory cosmetic value, and recurrence rates are also frequently reported (Fujimoto et al 2007, Neeli et al 2012).

Excision of the cyst

Excision of the cyst is the "Gold Standard" which has an almost unreported recurrence rate and a much better cosmetic result. This procedure is performed under general anesthesia, but in cooperative children it can be performed under local anesthesia (Heather et al 2011).

Whereas a study conducted by Fujimoto et al., 2010, which discuss about parameatal cysts in 5 neonatal girls. The cyst finally disappear spontaneously within 76-304 days after birth (Fujimoto et al 2007).

Histologically, there are 4 types of epithelium that can line the cyst wall, there are columnar, cuboidal, squamous, or transitional epithelium which varies according to the origin of the lesion in the urethra. Otsuka et al. describe the three histopathological classifications of cysts into 3 types, there are epithelium which origin from urethra (columnar, cuboidal, and/or transitional), followed by epithelium which origin from epidermis (squamous epithelium), and mixed type (Gupta & Gupta 2015, Lal & Agarwal 2013, Willis et al 2011, Herek et al 2000).

The largest retrospective study conducted by Willis et al. (2011), on 18 prepubertal males, by looking at demographic data, symptoms, pathology, cyst symptoms, and treatment that had been done. From the data reported, almost all of these cysts are asymptomatic, but 1/3 of the patients complained of pain when urinating, and urinary branching. The incidence of this disease in 12 boys, reportedly found at birth or 1 year after birth, and 6 patients, were unaware of the initial perimeatal cyst of the urethra (Willis et al, 2011). Parameatal cysts may be located in the urethra which is located laterally, or in the urethral ventral meatus. One patient reported with bilateral cysts. The diameter measured was less than 1 cm, 12 persons with a diameter of 5 mm or less. Fifty percent of patients had previously undergone circumcision, and 1/3 did not report data. Fourteen patients who underwent excision of the cyst, none had reported relapse and other complications. Four patients who did not undergo surgery, 1 experienced spontaneous resolution within weeks, while the other 3 were under observation and did not seek other treatment (Gupta & Gupta 2015, Willis et al 2011, Elder 2007). The histopathologic results reported in this study, apart from the specimens reviewed by Willis et al (2011), consist of a combination of epithelium (transitional epithelium, cuboidal, squamous and columnar epithelium). Three patients reported 3 single epithelium, namely transitional/cuboidal/columnar epithelium. The transitional and cuboid epithelium are the most frequently reported epithelium, followed by the columnar and squamous epithelium. Elder et al. described the parameatal cyst parameter of the urethra consisting of transitional, squamous or columnar epithelium. None of the specimens obtained were malignant and only 1 showed inflammation (Gupta & Gupta 2015, Willes et al 2011, Elder 2007).

CONCLUSION

Parameatal cyst of the urethra is an abnormal and rare case. Often without symptoms, but several symptoms such as pain when urinating, branching urine, to urinary retention are also reported. Physical examination alone can be used to make a diagnosis. Based on histopathological results, these cysts are rarely associated with infection or inflammation and may consist of several types of epithelium in them. Parameatal urethral cysts may disappear spontaneously in neonates, but there is a risk of recurrence. Complete surgical excision has been reported to have good cosmetic results without relapse.

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