RESEARCH ARTICLE / *Araştırma Makalesi*

Seminal Vesicle Cyst with or without Unilateral Renal Agenesis: Diagnosis and Treatment Options

Tek Taraflı Renal Agenezili veya Agenezisiz Seminal Vezikül Kisti: Tanı ve Tedavi Seçenekleri

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Abstract

Objective	The aim of this study is to report our experience on the management of seminal vesicle cyst which is an exceedingly rare acquired or congenital pathology.
Materials and Methods	A retrospective review was conducted on seven patients diagnosed with seminal vesicle cyst between March 2011 to March 2020. Patients' complaints, fertility status, physical signs, diagnostic tests or intervention and the obtained findings, surgical intervention, duration of follow-up period, complications, recurrence and histologic examination of the cysts wall were evaluated in patients included.
Results	Seven male patients, mean aged 36.29±13.45, IPSS 13.57±4.89, and Qmax:21.77±3.42ml/s with seminal vesicle cysts were identified. The patients' complaints included lower abdominal pain in one patient with giant seminal vesicle cyst and perineal pain, and storage lower urinary tract symptoms in six patient. Five patients were fertile and semen analysis was within normal ranges, two patients were infertile. Six patients have ipsilateral renal agenesia. One asymptomatic patient, and three patients with storage lower urinary tract symptoms followed without intervention. Two infertile patients were operated with transurethral resection. The cysts of the seminal vesicles >12cm were evaluated as giant cysts, and operated by open excision. Neither complications nor recurrences were observed. Histopathologic examination of the samples were reported to be compatible with seminal vesicle cyst.
Conclusion	The open surgical approach might be considered the definitive form of treatment for giant cysts. Although small asymptomatic seminal vesical cyst can be followed without intervention, the symptomatic cyst protruding to bladder can be managed by transurethral route.
Keywords	Infertility; renal agenesis; seminal vesicle cyst.

Öz

Amaç	*Son derece nadiren veya doğuştan bir patoloji olan seminal vezikül kistinin yönetimi hakkındaki tecrübemizi bildirmektir.*
Gereç ve Yöntemle	*Kliniğimizde Mart 2011-2020 yılları arasında seminal vezikül kisti tanısı alan yedi hasta retrospektif olarak incelendi. Hastaların şikayeti, fertilite durumu, fizik muayene bulguları, tanısal testleri, görüntülemesi ve elde edilen bulguları, cerrahi türü, takip süresi, komplikasyonlar ve rekürens varlığı, kist duvarının histolojik incelemesi kaydedildi.*
Bulgular	*Seminal vezikül kisti saptanan yedi erkek hastanın, ortalama yaş 36.29 ± 13.45, IPSS 13.57 ± 4.89, Qmax: 21.77 ± 3.42ml/sn idi. Dev seminal vezikül kisti olan hastada perineal ağrı ve alt karın ağrısı, altı hastada depolama alt üriner system semptom şikayetleri mevcuttu. Beş hasta infertil ve semen analizi normal aralıklardaydı, iki hasta infertildi. Altı hastada ipsilateral renal agenezi, bir asemptomatik hasta ve üç hasta depolama alt üriner sistem semptomları tedavisiz takip edildi. İki infertil hasta transüretral rezeksiyon ile opera edildi. Seminal vezikül kisti> 12 cm dev kistler olarak değerlendiridli ve açık eksizyon uygulandı. Komplikasyon veya nüks gözlenmedi. Numunelerinin histopatolojik incelemesinin seminal vezikül kisti ile uyumlu olduğu bildirildi.*
Sonuç	*Açık cerrahi yaklaşım, dev kistler için kesin tedavi şekli olarak kabul edilebilir. Her ne kadar küçük asemptomatik seminal vezikal kistin müdahale olmadan takip edilebilse de, mesaneye protrüde semptomatik kist transüretral yolla tedavi edilebilir.*
Anahtar Kelimeler	*İnfertilite; renal agenezi; seminal vezikül kisti.*

INTRODUCTION

Seminal vesicles are a pair of saccular organs that are located at the posterior of the bladder and that lie between the bladder and the rectum. The seminal vesicle has the volume for 3 to 4 mL and the nonobstructed seminal vesicle usually measures 5 to 7 cm in length and 1.5 cm in width.[1] Primary pathologies related to seminal vesicles are rare but the advances in the imaging technologies has been increasing their incidence. Seminal vesicle cysts are seen infrequently with an incidence of 0.005%,[2] generally asymptomatic and are detected coincidentally. The vast majority of seminal vesicle cysts are asymptomatic. Congenital seminal vesicle cysts are generally related to abnormalities of the mesonephric (Wolffian) duct, and the acquired cysts usually consequence from ejaculatory duct obstruction as further sequelae of infection, urethral inflammation, or ejaculatory duct stones.[3] Hematospermia, terminal hematuria, lower abdominal or lumbosacral pain, testicular and perineal discomfort may observed with seminal vesicle cysts. Symptomatic cysts may also result in obstruction-related hydronephrosis and problems with urination or defecation. They are generally associated with ipsilateral renal agenesis and ectopic ureter and require surgical management.[3]

Zinner syndrome is a rare congenital malformation of the urogenital system and is characterized by the triad of seminal vesicle cyst, ejaculatory duct obstruction and ipsilateral renal agenesis. Less than 200 cases have been reported in the literature.[4] In our study, seven patients had seminal vesicle cysts and six patients had unilateral renal agenesis. Seminal vesicle cysts are generally uncovered incidentally in the second and fourth decades, when sexual and reproductive activity is at its highest. In this study we retrospectively evaluated our patients with isolated seminal vesical cysts and associated renal agenesis who were treated with different surgical techniques and we compared our results with those of previous studies.

MATERIALS and METHODS

A retrospective chart review was conducted on seven adult men who presented to the urology outpatient clinic of Sakarya University hospital from March 2011-March 2020 were diagnosed with seminal vesicle cyst, and subsequently treated. Patients' complaints, fertility status, physical signs, diagnostic tests or intervention and the obtained findings, surgical intervention, duration of follow-up period, complications, recurrence and histologic examination of the cysts wall were evaluated in patients included. Local institutional review board (Sakarya University) approval (date: 05.06.2020, study no. 71522473/050.01.04/330) was obtained before study initiation. The study followed the ethical principles of the Declaration of Helsinki. Data obtained in this study were evaluated and statistically analyzed using SPSS (SPSS version 21.0, IBM, Armonk, NY, USA) package software. General features of the study population were expressed using descriptive statistics. The quantitative variables were expressed as mean ± standard deviation, and minimum, maximum. The qualitative variables were given as number and percentage.

RESULTS

One patient presenting to our Urology outpatient clinics with left lower abdominal pain and 6 patients (85.7%) presenting with perineal pain, storage predominant lower urinary tract symptoms were diagnosed with seminal vesical cysts after urinary ultrasonography and computed tomography (CT) of abdomen. The last patient with right solitary kidney referred to our outpatient clinic by Nefrology clinic due to right seminal vesicle cyst without any symptom.

The mean age was 36.29±13.45 (min-max: 21-63) years, mean IPSS was 13.57±4.89 (min-max: 6-19), mean Qmax was 21.77±3.42 ml/s (min-max: 17.1-27.7), mean voiding volume was 254±29 ml (min-max:180-330ml). All patients' scrotal examinations were within normal range. On digital rectal examination of these patients a fluctuating compressible mass originating from the superior portion of the prostate gland was palpable. 29-year-old man with

symptoms of perineal pain, and LUTS storage dominant, the axial section of the seminal vesicle cyst in CT is shown in Figure 1. All patients were examined with transrectal ultrasound, routine blood tests, urine analysis, culture and uroflowmetry. All patients' IPSS scores were recorded. The routine blood tests and urine analysis of all patients revealed normal findings. Renal agenesis were detected on CT of six patients (85.7%) (Figure 2). In cystoscopic assessment of four of these six patients, the left ureteric orifice and hemitrigon were not observed. Cystoscopy was not performed in 2 patients.

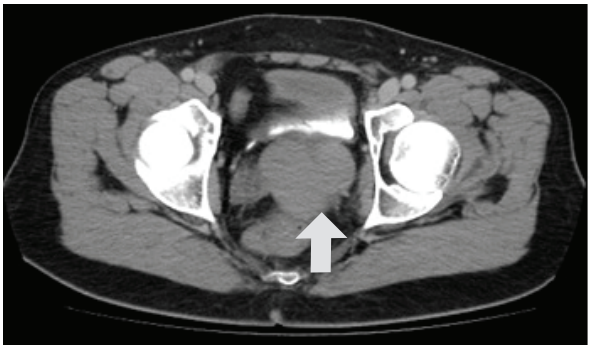

Figure 1. 29-year-old man with symptoms of perineal pain, and LUTS storage dominant.
Axial unenhanced CT scan shows large seminal vesicle cyst (arrow) to be well-defined low attenuation mass posterior to bladder and arising from seminal vesicle.

Figure 2. 28-year-old man with symptoms of perineal pain, and LUTS storage dominant.
Coronal reformation shows associated ipsilateral agenesis of left kidney. Normal right kidney is visible. Again note presence of left seminal vesicle cyst (arrow).

The biggest dimension of the cyst were 20x25 cm in one patient which may be called a giant cyst. The predominant symptom of the patient with giant cyst was ipsilateral lower quadrant pain. Also ipsilateral renal agenesis was detected in this patient. This patient was fertile and his semen analysis parametres were normal. The cyst of this patient was resected with a infraumbilical median incision without opening the peritoneum (Figure 3).

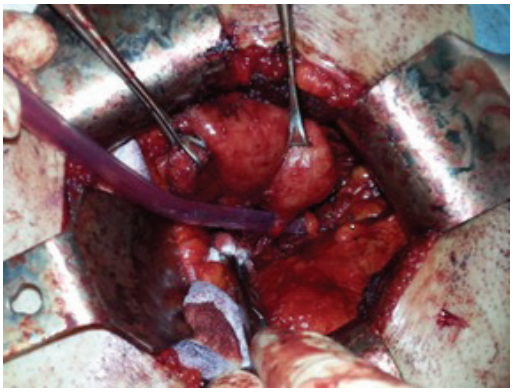

Figure 3. Surgical view of the seminal vesical cyst (asteriks) (Consent was obtained from the patient to publish this image).

The predominant symptom of the second and third patients were filling lower urinary tract symptoms and perineal pain. In one of these patients' ipsilateral renal agenesis, and two of them azoospermia were detected. These symptomatic two patients' seminal vesicle cysts were proximal, adjacent to the prostate, transurethral resection was performed to unroof the cysts at the 5-o'clock and 7-o'clock positions distal to the bladder neck.

The semen analysis parameters of the other four patients were normal. Fourth, fifth, sixth patients' storage predominant lower urinary symptoms were subsided with anticholinergic and anti-inflammatory drug use. They have not had any surgical intervention during follow-up period. Seminal vesicle size did not change significantly in US during a mean follow-up of twenty-four months.

The mean operative time was 32 minutes for the patients who underwent transurethral resection and 58 minutes for the open resection. The presenting symptoms were completely relieved and no postoperative complications were detected. Histopathologic examination of the samples were reported to be compatible with seminal vesicle cyst. The postoperative semen analysis of the patient presenting with azoospermia and to whom transurethral unroofing was performed eventually, was positive for sperms in postoperative third-months. Although no increase in seminal vesicle cyst diameter was detected in the ninety-six months follow-up period, he presented to our outpatient clinic with storage-predominate lower urinary tract symptoms. The patient is still on alpha blocker and anticholinergic therapy. The patient's serum creatinine level did not change. Patient characteristics are shown in Table 1.

Table 1. Patient Characterics

	Patient 1	**Patient 2**	**Patient 3**	**Patient 4**	**Patient 5**	**Patient 6**	**Patient 7**
Age (year)	39	21	28	29	39	35	63
Cyst size (cm)	20x25	4x5	4x5	3x3	4x2	4x3	6x5
Chief complaint	Perineal pain, LUTS storage dominant, dull pain at left lower quadrant	Perineal pain, LUTS storage dominant	Perineal pain, LUTS storage dominant	Perineal pain, LUTS storage dominant	Perineal pain, LUTS storage dominant	Perineal pain, LUTS storage dominant	Asymptomatic
Total IPSS	6	16	17	19	17	9	11
Qmax (ml/s)	19.5	21.9	20.4	27.7	21.5	24.3	17.1
Voiding volume (ml)	205	270	310	330	235	250	180
Presence of renal Agenesia	Yes	No	Yes	Yes	Yes	Yes	Yes
Fertility status	Yes	No	No	Yes	Yes	Yes	Yes
Semen analysis	Normospermia	Azospermia	Azospermia	Normospermia	Normospermia	Normospermia	Normospermia
Surgery	Open resection	TUR	TUR	N/A	N/A	N/A	N/A
Operation time (min)	58	34	30	N/A	N/A	N/A	N/A
Post-operative complication	No	No	No	N/A	N/A	N/A	N/A
Follow-up time (months)	108	108	120	24	24	30	96
Supplementary note	N/A	Postoperative third-months semen analysis was normospermia.	Postoperative third-months semen analysis was normospermia.	N/A	N/A	N/A	N/A

IPSS: International Prostate Symptom Score, LUTS: Lower urinary tract symptoms TUR: Transurethral resection

DISCUSSION

Human seminal vesicle appears as a diverticulum of the Wolfian duct at about the 10th to the 12th week of gestation. The diverticulum elongates and folds back on itself, thus forming a "hooklike duct" with extensive "side ducts forming during months 4 to 5 of pregnancy".[5] Seminal vesicles fuse with the ampulla of the vas deferens to form the ejaculatory ducts that empty into the prostatic urethra via the verumontanum.[3] The seminal vesicles are paired male reproductive organs that produce about 60% of the seminal fluid and they are located posteriorly to the prostate.[6]

The seminal vesicle contractible, compliant, smooth muscular organ with dynamic properties analogous to those of the bladder, and the ejaculatory duct serves as a urethra-like conduit. This theory allows the classification of ejaculatory duct obstruction into two types of disorders, analogous to bladder outlet obstruction. First, obstruction resulting from physical blockage of the ducts, similar to bladder outlet obstruction. Second, functional obstruction of the seminal vesicle, similar to voiding dysfunction caused by bladder myopathy. As a result, this unit is analogous to the bladder and urethra and is exposed to physical blockage and functional disorganizations that may result in infertility.[7]

Unilateral renal agenesis is a condition in which an individual is born with a nonectopic, solitary functioning kidney and failure of the contralateral kidney to develop. Also, it occurs in 1 in 1200 live births. Complete absence of unilateral renal agenesis observed more often than bilateral renal agenesis but is not easily detected on physical examination and is ordinarily an coincidental finding during an imaging study.[8]

Zinner syndrome is a seminal vesicle cyst caused by obstruction of the ejaculatory duct can be seen in association with ipsilateral renal agenesis. In cases of seminal vesicle cysts and unilateral renal agenesis, the ureter may insert into the prostatic urethra or seminal vesicle. The diagnosis of unilateral renal agenesis should be suspected when the vas deferens or body and tail of the epididymis are not palpable when in males evaluated for infertility.

Congenital seminal vesicle cysts may co-exist with urinary malformations such as ipsilateral renal agenesis, dysplasia, ectopic ureters and ipsilateral absence of the testicle, cryptorchidia and hypospadias which is due to the very close location of ureteric bud and the mesonephric canal during embryogenesis. Cysts are generally lined with cuboidal or squamous epithelium which is filled with a clear liquid and all of them are encapsulated in a fibrous capsule.[9]

Acquired cysts are generally unilateral. These are generally a result of infection and obstruction of ejaculatory ducts and seminal vesicles secondary to prostatitis or prostatic surgery. This is frequently secondary to a retrograde urinary infection. Acquired cysts are associated with benign prostatic enlargement, prostatic surgery and malignancy. They can be round or oval shaped and more infrequently they can be tubular and multilocular.[10] Since seminal vesicle cysts are androgen dependent and they mature in the puberty they generally become symptomatic at the ages of 20-30 years with a mass effect.[11] This presentation is thought to be a result of the cystic dilatation after a collection of liquid in seminal vesicles.[10] Symptoms are a result of the pressure exerted on the other organs by the dilated cyst or it's irritative effect. Generally those cysts over 5 cm in diameter are symptomatic and require therapy.[12]

Voiding and storage lower urinary tract symptoms and hematuria can occur if the cyst is in the neighborhood of bladder. Perineal and suprapubic pain, hypogastric pain and problems of defecation can occur depending on the size of the cyst. Obstruction of spermatic canals can cause hematospermia, postcoital pain, epididymitis and infertility. Most frequent symptoms are storage lower urinary tract symptoms, postcoital pain and epididymitis.[13] Bigger cysts may cause rectal obstruction that present as tenesmus or constipation.[13-15]

Seminal vesicle cysts <5 cm can remain asymptomatic and these are generally diagnosed incidentally upon digital rectal examination as fluctuating masses originating from the superior surface of the prostate gland. These cysts may manifest as bladder irritation and obstruction related symptoms.[16]

Seminal vesicle cysts larger than 12 cm are evaluated as giant cysts. Frequently they manifest as bladder and colonic obstruction symptoms.[12] Giant cysts cause voiding difficulty by inducing bladder irritation and obstruction. Nevertheless in our patient presenting with a giant cyst the only complaint was abdominal pain located in the lower left quadrant and he was fertile with normal semen analysis parameters. In the literature the most commonly reported symptoms were abdominal, perineal and pelvic pain, painful ejaculation, dysuria, pollakuria, urinary tract infections, hematuria, epididymitis and signs of prostatitis. Also cases presenting with infertility, hematospermia and enuresis were reported.[14,15]

The initial diagnostic evaluation of the giant seminal vesicle cyst are abdominal and transrectal ultrasonography. Abdominal CT scan, magnetic resonance imaging and seminal vesiculography may also be useful in detecting co-existing deformities. The seminal vesicle cyst appears in CT scan as a water dense, well demarcated cystic mass originating from the seminal vesicle and located in the rectovesical space. Cystoscopic evaluation may also be useful in determination of other anomalies of hemitrigon, absence of ureteral orifice and other anomalies of the bladder. The routine laboratory findings are generally normal. Culture of seminal fluid or prostatic secretions collected after prostatic massage may be positive for an infection. Chronic infection may also compromise sperm parameters causing infertility.[12,15] Seminal vesiculography may be useful in selected cases in detecting other pathologies such as Wolff or Mullerian canal cysts, ejaculatory duct diverticuli, prostatic cysts, Dougles abcess, leiomyoma and leiomyosarcoma of the bladder and ureterocele.[12] The differential diagnosis primarily includes cystadenoma and papillary adenoma which are benign tumours of the seminal vesicles, Mullerian canal cysts, ectopic ureterocele, diverticuli of ejaculatory ducts or ampullary region of the vas deferens, prostatic cysts, primary (adenocarcinoma and sarcoma) and secondary malignant tumours of seminal vesicles that are extentions of the primary tumours of bladder, prostate or rectum and lymphoma. In the male the differential diagnosis of the cystic pelvic masses include Mullerian duct cysts (prostatic utricle) and Wolfian duct cysts (seminal vesicles, vas deference or ejaculatory duct cysts).[11]

The transrectal or transperineal drainage of the cyst is not preferred due to high risk of infection and high frequency of recurrence after drainage.[12] In literature transurethral resection/incision of the anterior wall of the cyst intravesically has been reported.[17] This procedure is associated with a risk of potential infections and abnormalities in the semen analysis parameters due to contamination of the system by urine. Nevertheless two cases with cyst dimentions of 5x6 cm and with complaints of painful ejaculation and perineal pain were treated with transurethral resection. In small cysts finding the cyst, during open surgery is reported to be challenging. In our cases transurethral unroofing of the cysts were performed because abdominal CT revealed that the cysts were bulging into the bladder which made them easily accessible through bladder, and the operations were performed without any complications. The patient with azoospermia and renal agenesis recovered completely at postoperative month 6 except symptoms of mild lower urinary tract symptoms and his semen analysis was positive for sperms. The other patient with a cyst of 20 cm was underwent open surgery due to the great dimensions of his cyst. This patient was operated under general anesthesia with an infraumblical midline incision. No perioperative or early postoperative complications occurred.
Limitations of the study

The limitations of our present study include its retrospective nature and the relatively small number of patients studied.

CONCLUSIONS

In recent years as a consequence of widespread use of laparoscopic techniques in urology, laparoscopic cyst excision has become one of the treatment alternatives.[18,19] Besides robot assisted laparoscopic techniques has also shown feasibility and effectiveness in treatment of cysts. Also in centers without an adequate level of experience in laparoscopic techniques or without proper equipment for robotics these cysts can be successfully removed with open surgery without a need of opening the peritoneum. Small cysts can be approached via transurethral route and unroofing can be performed.

Ethics statement: The present study protocol was reviewed and approved by the Institutional Review Board of Sakarya University College of Medicine (approval date: date: 05.06.2020, study no. 71522473/050.01.04/330). Informed consent was obtained by all subjects when they were enrolled.

Abbreviations

CT: computed tomography

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

The project was developed by OK. The clinical database of the patients was acquired by OK, YTA, DG, HIC. The manuscript was written by OK, AE. The operative procedures were performed by OK.

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