Epididimal Cyst in Children: A Single-Institutional Experience

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Purpose: Simple epididymal cysts (EC) are rare in childhood and are mostly diagnosed at puberty. Although there is no consensus on the treatment, a conservative approach is generally preferred. To evaluate patients diagnosed with EC at our clinic in terms of presenting symptoms, diagnosis, and treatment methods.

Materials and Methods: Data of patients treated for epididymal cysts at our institution between March 2012 and March 2023 were retrospectively analyzed in terms of age, symptomatology, diagnostic method, treatment method, and outcomes. In all cases, the diagnosis of EC was based on physical examination with scrotal ultrasonography (US) confirmation.

Results: A total of 1829 patients underwent scrotal Doppler US, and EC was detected in 72 patients (10.7%). The median follow-up period of the 43 patients was 21.7 (6-80 months). Of these, 9 were bilateral (12.5%). The mean age of the patients at presentation was 14.8 years. Forty-one patients had scrotal pain, 12 had scrotal swelling, and 19 incidentally had EC. The cysts were between 1.2- 37 mm. Only 3 (4.1%) patients required surgical excision due to persistent pain.

Conclusion: EC is a benign lesion, and treatment approaches are usually conservative. Surgical excision is recommended for patients with persistent scrotal pain or an acute scrotum.

Keywords: children; epididymal cyst; management; surgery; ultrasonography

INTRODUCTION

Unlike intratesticular masses, most extratesticular masses are benign. The most common extratesticular lesions in children include Morgagni-hydatid torsion, epididymitis, paratesticular rhabdomyosarcoma, epididymal cysts/spermatoceles, and varicocele. In children and adolescents, EC are benign lesions that are usually diagnosed by US during the evaluation of scrotal pain or masses. An EC is a dilatation of the efferent epididymal tubules due to tubular obstruction and fluid accumulation in a single (unilocular) or multiple (multilocular) sacs. They usually originate in the head of the epididymis. Cysts arising from the tail of the epididymis are rare[(1)]. Different EC rates, ranging from 5% to 20%, have been reported in the literature[(2)]. This is more common in the pubertal age group than previously thought. Up to 20% of cases may involve bilateral or multiple cysts in a single epididymis[(3)]. EC are benign cystic lesions containing serous fluid in the epididymis and are lined with smooth columnar epithelium, unlike spermatoceles, which contain sperm upon aspiration[(4)].

EC has been reported to be common in children with cryptorchidism, cystic fibrosis, polycystic kidney disease and von Hippel-Lindau syndrome. In addition, ipsilateral renal agenesis, ejaculatory system atresia, and ipsilateral hemitrigoneal absence have been described, although they are extremely rare[(5-8)].

Although scrotal pain is a common complaint in EC, it may go unrecognized when the cyst is small and is usually asymptomatic. 70-80% of cases are detected incidentally during scrotal US performed for another reason. However, 20-30% of cases are detected during physical examination and confirmed with scrotal US[(9-11)].

EC are benign, and often self-limiting, and their treatment is usually conservative. Most cases (up to 60%) regress spontaneously, especially if they are less than 1 cm in size[(10,12)]. Although some publications recommend follow-up below 3 cm, cysts> 1 cm should be well differentiated from other pathologies that require surgical treatment, as the risk of torsion increases.

MATERIALS AND METHODS

Medical records, scrotal US findings, and Doppler US results of all patients diagnosed with EC between March 2012 and March 2023 were reviewed. Age, clinical course, concomitant anomalies and diseases, cyst and testicular dimensions, time-to-cyst regression, treatment modality, and outcomes were retrospectively analyzed. The inclusion criteria were age < 18 years and access to clinical and US records. The exclusion criterion was a patient who was followed up once during the file review.

All statistical analyses were performed using SPSS 20.0 (IBM Corp., Armonk, NY, USA). Shapiro-Wilk's test

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Table 1. Baseline characteristics of the patients

Variable	Group 1 (2-48 m)	Group 2 (10-17 y)	Total
Age	12 (27.9%)	31 (72.1%) 43	
Side			
Left	4	11	15 (34,8%)
Regresion	2	0	2 (13,3%)
Right	5	14	19 (44,8%)
Regresion	2	3	5 (26,3%)
Bilateral	3	6	9 (20,9%)
Regresion	1	0	1 (11,1%)
Regresion total	5 (41,6%)	3 (9,6%)	8 (18,6%)
Cyst size	1-20 mm	1.2-29 mm	
Genitourinary anomalies	3	2	
İnguinoscrotal diseases	6	18	

Abbreviations: m, month; y, year

was used to assess the assumption of normality. Continuous variables were presented with median (interquartile range (IQR)). Comparisons of continuous variables between groups were carried out using the Mann-Whitney U test. Dependent group comparisons were performed by the Wilcoxon signed rank test. A *p*-value <.05 was considered significant.

This study was conducted in accordance with the Kocaeli University Ethics Committee Decision: E-80418770-020-413327 date and number, 2023/163 project number.

RESULTS

EC was detected in 72 (7.68%) of 937 children who underwent scrotal Doppler US between March 2012 and March 2023. Nine (12.5 %) patients had bilateral lesions. Fifty-three patients presented with scrotal pain and/or swelling (73.6%) and 19 patients had EC incidentally (26.3%). The mean patient age at presentation was 14.8 years (2 months-17 years). It was observed that the ages of the patients were divided into two age groups: under 4 years old and over 10 years old. No patient was between the ages of 4 and 10 years. Therefore, the patients were divided into two groups, taking these age groups into consideration the younger group aged 2 months-4 years (n = 17) and the pubertal age group aged between 10-17 years (n = 55).

The study group consisted of 43 patients with a median follow-up period of 21.7 months (range, 2–80 months). The rate of EC increases with age and is most common during puberty (72%). In the study group, 19 patients had right EC (44.1 %), 15 had left EC (34.8 %), and 9 had bilateral EC (20.9 %) (**Table 1**).

The size of the cysts ranged from 1.2 to 37 mm, with a median size of 7.4 mm. There were 31 patients (72.1%) with a cyst size between 1,2-9 mm and 12 patients (27.9%) with a cyst size between 10-37 mm. In patients with unilateral EC, there was no significant difference between the mean size of the same-side testes and the mean size of the contralateral testes (p = .986) (**Table 2, Figure 1**).

Surgical excision was performed in three patients (4.1%) aged 13-17 years due to persistent scrotal pain. One patient who underwent surgery developed a cyst in the opposite epididymis one year later. Spontaneous regression was observed in eight (18.6%) patients with EC. One patient experienced had bilateral regression, and the mean duration was 23 months. The regression rate was 41.6% in the young age group (2 months-4 years, 12/5) and 9.6% in the pubertal group (10-17 years, 31/3).

Genitourinary anomalies (n = 5) and inguinoscrotal diseases (n = 24) accompanied by EC were also observed (**Table 3**). None of our patients had a history of exposure to diethylstilbestrol, cystic fibrosis, or von Hippel-Lindau disease.

DISCUSSION

Although EC is extremely rare in childhood, its incidence increases with age and is attributed to hormonal conditions[11]. The incidence of EC in children varies between 5%-20%, and the incidence rate of EC in this study (7.68%) was consistent with that reported in the literature[2]. They present as single or multiple unilateral or bilateral spherical cysts, localized to the head of the epididymis. In our study, the bilateral incidence rate (20.9%) did not different from that reported in the literature[10,12].

There is no consensus on the pathogenesis of EC, and various theories have been proposed. Although EC is widely considered an acquired condition, some authors have suggest that it could be associated with a syndrome of testicular dysgenesis caused by various endocrine disruptors acting during embryo growth and development[13,14]. Wollin et al. defined an EC as a structure arising from the remnants of the epididymis that is not connected to the epididymal tubules[15]. It is unknown whether these remnants are of a mesonephric or Mullerian origin. McLachlan et al. suggested that EC result-

Table 2. Comparison of PCa, CsPCa detection rate and CsPCa proportion between the two groups.

Variable	N	M 1	M 2	P value	95% CI
Testes				.986	-4.07 - 3.98
Semi testes	43	10.8554	8.7470		
Contralateral testes	41	10.9033	7.6540		

Abbreviations: N, number; M 1, Mean; M 2, Median; CI,confidence interval

Table 3. Genitourinary anomalies

Comorbidities	Patient count	Age
Hydrocele and/or İnguinal hernia	9	1m-17y
Varicocele and Nutcracker Syndrome	4	16-17y
Epididymorchitis	3	12-17y
Testicular torsion	2	15-17y
Undescended testis	3	3-15y
Bilateral Microlithiasis	2	13y
Hydronephrosis (grade-3)	2	1m-1y
Ambigius genitale+Hypospadias	1	4y
Mixt Malignant Germ Cell Testicular Tumor	1	15y
Kidney cyst	1	16y
Renal agenesis	1	5 m
Total	29 (67.4%)	

Abbreviations: m, month; y, year

ing from fetal estrogen exposure can be identified as Mullerian (paramezonephric) rather than mesonephric by detecting lactotransferrin, an estrogen-dependent protein found only in Mullerian tissue[16]. An increased incidence of EC has been observed in boys exposed to diethylstilbestrol during fetal life[9,10,12]. It has also been reported to occur after hormonal disturbances, degenerative processes, and obliteration or stenosis of the epididymal ducts, resulting from toxic agents acting after birth[5,17]. The pathogenic mechanisms of EC have not yet been fully elucidated and more extensive studies are needed.

EC has been found to be associated with cryptorchidism, cystic fibrosis, von Hippel-Lindau disease[5-7]. Congenital cystic dysplasia, a new congenital epididymal lesion, has recently been associated with renal and/or urinary tract malformations. The authors envisaged a common pathogenic mechanism characterized by impaired mesonephric duct growth with disruption of the basic molecular pathways[18]. In our series, renal and/or urinary system malformations were observed in 11.6% of cases, and simple renal cysts, renal agenesis, and hydronephrosis were observed in association with EC. Patients diagnosed with grade-3 hydronephrosis had EC on the same side. None of the patientS required surgical intervention, and hydronephrosis regressed in one-year follow-up. This finding supports the definition of "congenital cystic dysplasia" proposed by Nistal et al. Urinary system screening was performed in 41.8% of the patients. We believe that urinary sonography and screening of the urinary system, particularly the kidneys, should be routinely performed in patients with EC.

The incidence rate of inguinal disease accompanying EC was 55.8% (**Table 1**). The most common accompanying pathologies in our patients with EC were hydrocele and/or inguinal hernia, varicocele, epididymo-orchitis, and undescended testes, consistent with the literature. Two patients underwent surgery for testicular torsion and had multiple cysts on the same side. Avsarlar et al. predicted that an increased number of these cysts is associated with a higher risk of testicular atrophy[19].

Approximately 20-30% of EC are identified on physical examination and confirmed by US scanning of the scrotum. US helps determine the location and size of the cyst within the epididymis. It is most commonly observed in the head of the epididymis and more rarely in the body and tail[20,21]. Sonographically, the EC appeared as a simple, echo-free cystic structure (**Figure 1**).

Painful or painless scrotal edema or swelling may explain this presentation. While 51.8-75% of cysts are asymptomatic and found incidentally during physical examination, 25-49.2% are symptomatic[9-11,14]. Among our patients, 73.6% had pain or scrotal swelling. Although EC are benign and often self-limiting, they should be well differentiated from epididymal torsion, testicular torsion, and other causes of acute scrotum that require urgent medical or surgical treatment[22]. To date, nine cases of EC torsion have been reported in the pediatric literature, all of which require surgical treatment[23]. Parents should be warned about an acute scrotum may develop in a patient following the diagnosis of EC.

Differential diagnoses should include epididymitis, orchitis, hydrocele, varicocele, extratesticular scrotal masses, epididymal or testicular tumors, complicated testicular or epididymal appendices, and spermatocele. Adenomatoid tumors are the most common benign lesion of the epididymis, and papillary cystadenomas occur in 25% of patients with von Hippel-Lindau syndrome[7]. Therefore, given the rare association with malformations or tumoral pathologies, physical examination, Doppler US, scrotoscopy, or surgery should not be avoided if necessary for complete diagnosis[4,21,22,24]. We also diagnosed a mixed malignant germ cell testicular tumor in one of our patients.

Different methods can be used to measure the testicular volume. One of these is US. More practically, although it is subjective and the results may vary according to the practitioner, the orchidometer developed by Prader, which consists of molds shaped like testes with a volume between 1-25 mL is frequently used in testicular volume measurement[25]. In our study, testicular dimensions were measured using scrotal US. No significant difference was observed between measurements of the normal testes in patients with unilateral EC ($p > .05$). However, the limiting factor of this study was whether the size of both testes was normal or large, as in the study by Ekici et al.[12]. In a study conducted by Akyüz et al., regional differences in testicular size were observed in Turkish children[26]. Welliver et al. found variability in both inter- and intra-observer testicular volume measurements by private urologic ultrasonographers; and in more than 25% of cases, a difference of more than 20% in testicular volume measured in the same testes was found[27]. Considering the endocrine effects, measurement methods, personal factors, and regional factors, further studies are needed to determine whether testicular size is affected in children with EC. Although there is no consensus on treatment, in the majority of cases, the EC regresses spontaneously over

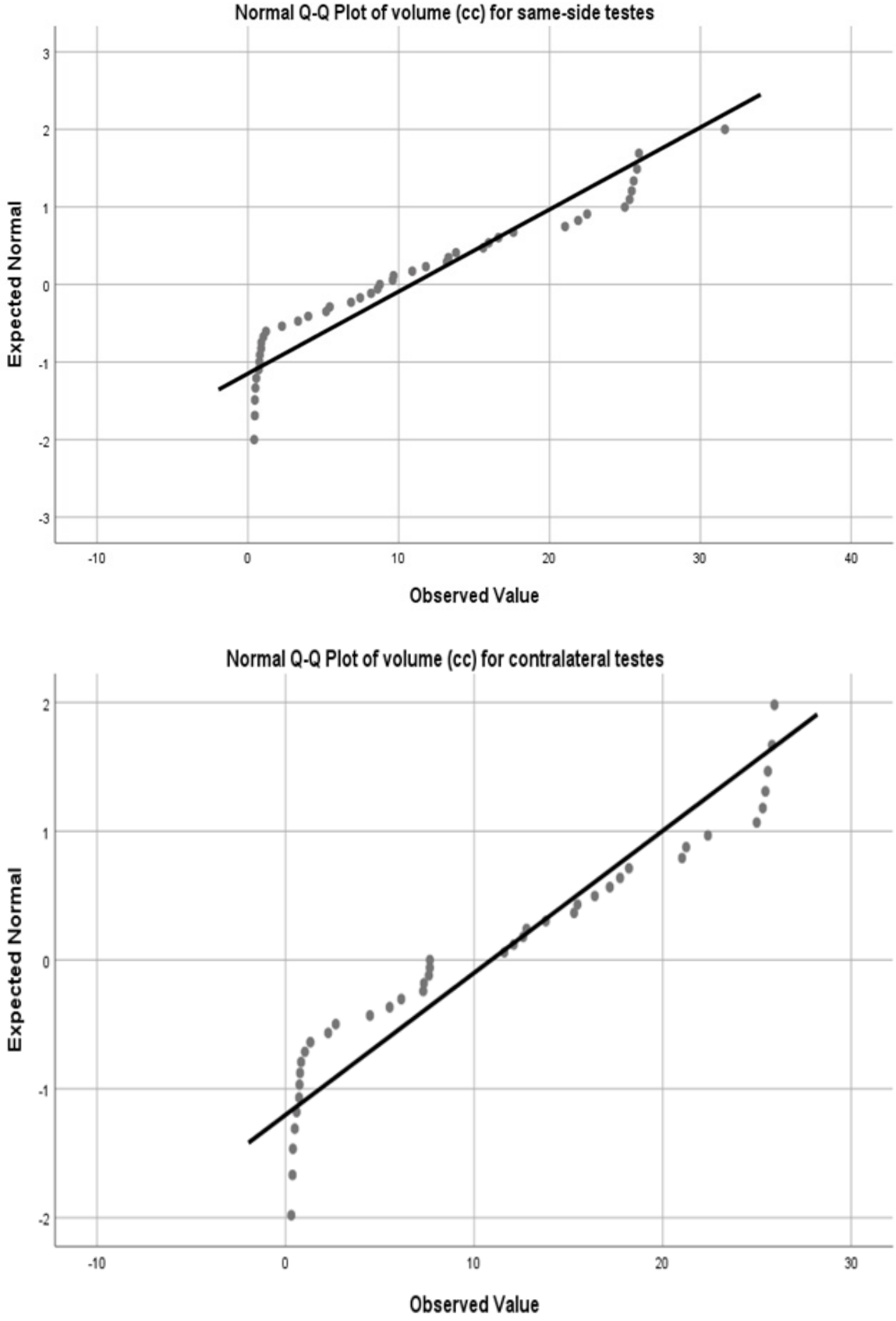

Figure 1 . Q-Q Plot of volume (cc) for same-side testes and contralateral testes

time and does not require surgical excision. There are no guidelines for the surgical intervention of EC. The recommended treatment method in childhood is an open surgical approach through a transverse scrotal incision or a scrotal raphe. Cyst size and pain play an important role in the choice of treatment. In the recent literature, the surgical rate varies between 5-52.5% and Homayoon et al. only one of 20 patients with EC in 12 years (5%), Erikci et al. 8 of 49 patients (19.1%) with EC in 3 years, Chillon et al. 6 of 15 patients (40%) with EC in 5 years, and Niedzielski et al. operated on 31 of 59 patients (52.5%) with EC in 6 years[10,12,28-30]. In our series, we operated on 3 of 43 patients (4.1%) over 11 years.

We limited our surgical criteria to a diameter> 1 cm and persistent pain. Some authors recommend surgery for cysts> 10 mm in diameter, whether symptomatic or asymptomatic, and others recommend surgical removal if the EC becomes symptomatic or begins to increase in size[9,10,12,29]. In 50% of patients with EC, cyst size regresses spontaneously within the next 3-35 months (mean, 18 months)[10]. Surgery should be limited to selected cases and possible complications should be avoided. Surgery should be avoided in young patients, especially in bilateral cases, because of the possibility of infertility. Surgery should be performed under microscopic vision in selected cases to prevent damage to the fine collecting tubules[9,10,12]. An alternative treatment option described in the literature is sclerotherapy using tetracycline, phenol, sodium tetradecyl sulfate, polidocanol, or ethanolamine oleate. However, injection of sclerotizing agents is not recommended in children[9,10]. Recently, surgical indications have been limited, and elective approaches have gained popularity. Our approach is close to follow-up if the pain complaint does not persist and emergency surgery is not required. In our study, the surgical rate was lower than that reported previously. Limiting surgery also reduces the risk of postoperative inflammation, infection, scrotal swelling, and infertility in older patients.

In the current literature, there is still no consensus on the follow-up and treatment of epididymal cysts in childhood. Our study has some limitations owing to its retrospective nature. Since some patients did not come for follow-up visits after diagnosis, follow-up data could not be obtained due to insufficient file data, and this patient group was excluded from the evaluation. However, it is valuable because it contains long-term data of 11 years of childhood epididymal cysts from a university hospital.

CONCLUSIONS

Epididimal cysts are benign lesions that are probably increasing in frequency, partycularly in pubertal boys. Conservative treatment results in the complete regression of most cysts. Surgery is recommended only in symptomatic patients or in cases of an acute scrotum. The fact that most patients are in adolescence limits the follow-up period to after 18 years of age. Longer-term studies whit an increased follow-up period are needed to determine the changes in cyst size and symptoms in adolescents after 18 years of age.

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CONFLICT OF INTEREST

The authors declare that they have no conflictsict of interest.

REFERENCES

1. Mukendi AM. Bilateral epididymal cyst with spontaneous resolution. Clin Case Rep. 2020;8:2689-91.
2. Rifkin MD, Kurtz AB, Goldberg BB. Epididymis examined by ultrasound. Correlation with pathology. Radiology. 1984 ;151:187-90.
3. Fernández-Ibieta M, Villalon-Ferrero F, Ramos-García JL. Benign Scrotal Tumor in a Pediatric Patient: Epididymal Cyst. Case Rep Urol. 2018;20;2018:1635635.
4. Munden MM and Trautwein LM: Scrotal pathol-ogy in pediatrics with sonographic imaging. CurrProbl Diagn Radiol. 2000;29:185.
5. Blau H, Freud E, Mussaffi H et al. Urogenital abnormalities in male children with cystic fibrosis. Arch Dis Child. 2002;87:135–8.
6. Belet U, Danaci M, Sarikaya S et al. Prevalence of epididymal, seminal vesicle, prostate, and testicular cysts in autosomal dominant polycystic kidney disease. Urology. 2002;60:138–41.
7. Joerger M, Koeberle D, Neumann HP, Gillessen S. Von Hippel-Lindau disease--a rare disease important to recognize. Onkologie. 2005;28:159–63.
8. Sheih CP, Hung CS, Wei CF, Lin CY. Cystic dilatations within the pelvis in patients with ipsilateral renal agenesis or dysplasia. J Urol 1990;144:324–7.
9. O'Kelly F, McAlpine K, Abdeen N et al. The futility of continued surveillance of epididymal cysts-a study of the prevalence and clinico-demographics in pre- vs. post-pubertal boys. Can Urol Assoc J. 2019;13:398–403.
10. Homayoon K, Suhre CD, Steinhardt GF. Epididymal cysts in children: natural history. J Urol. 2004;171:1274–6.
11. Posey ZQ, Ahn HJ, Junewick J, Chen JJ, Steinhardt GF Rate and associations of epididymal cysts on pediatric scrotal ultrasound. J Urol. 2010;184:1739–42.
12. Erikci V, Hoşgör M, Aksoy N et al. Management of epididymal cysts in childhood. J Pediatr Surg. 2013;48:2153-6.
13. Sharpe RM. Hormones and testis development and the possible adverse effects of environmental chemicals. Toxicol Lett. 2001;120:221-32.
14. Skakkebaek NE, Rajpert-De Meyts E, Main KM. Testicular dysgenesis syndrome: an increasingly common developmental disorder with environmental aspects. Hum Reprod. 2001;16:972-8.
15. Wollin M, Marshall FF, Fink MP, Malhotra R, Diamond DA. Aberrant epididymal tissue:

a significant clinical entity. J Urol. 1987 ;138:1247-50.
16. McLachlan JA, Newbold RR, Burow ME, Li SF. From malformations to molecular mechanisms in the male: three decades of research on endocrine disrupters. APMIS. 2001;109:263-72.
17. Patoulias D, Kalogirou M, Patoulias I. Intraparenchymal Epididymal Cyst (IEC) 4 cm in Diameter in a 15-Year Old Male Patient; a Case Report and Review of the Literature. Acta Medica (Hradec Kralove). 2016;59:137-9.
18. Nistal M, González-Peramato P, Sousa G et al. Cystic dysplasia of the epididymis: a disorder of mesonephric differentiation associated with renal maldevelopment. Virchows Arch. 2010;456:695-702.
19. Afsarlar CE, Elizondo R, Yilmaz E et al. Ultrasonographic findings in the epididymis of pediatric patients with testicular torsion. J Pediatr Urol. 2017;13:393.e1-6.
20. Arora BK, Arora R, Arora A. A randomised study of management modalities in epididymal cyst. Int Surg J. 2019;6:340-4.
21. Yin Z, Meng X. Chapter 4 -General techniques of scrotoscopic surgery. In: J Yang ed. Scrotoscopic Surgery. Cambridge, MA: Academic Press; 2019:23-53.
22. Karaman A, Afşarlar CE, Arda N. Epididymal cyst: not always a benign condition. Int J Urol. 2013;20:457-8.
23. Ozaal AMOM, Pragalathan B, Lavanya S, Sarma ST. Torsion of an Epididymal Cyst: A Rare Finding on Scrotal Exploration for Acute Scrotum. Case Rep Urol. 2020;22;2020:8858606.
24. Qin C, Yang J, Zhang R et al. The Application of Scrotoscope-Assisted Minimally Invasive Excision for Epididymal Mass: An Initial Report. Front Surg. 2022;24;9:804803.
25. Prader A. Testicular size: assessment and clinical importance. Triangle. 1966;7:240-3.
26. Akyüz O, Ergün M, Kılıç B et al. Testicular volume, and penile length of children aged 0-10 years in Turkey: Do measurements vary among geographical regions?. Androl Bul 2021;23:1−6
27. Welliver C, Cardona-Grau D, Elebyjian L, Feustel PJ, Kogan BA. Surprising interobserver and intra-observer variability in pediatric testicular ultrasound volumes. J Pediatr Urol. 2019;15:386.e1-6.
28. Boscarelli A, Bellini T. Epididymal cyst in children. Eur J Pediatr. 2021;180:2723-9.
29. Niedzielski J, Miodek M, Krakós M. Epididymal cysts in childhood - conservative or surgical approach? Pol Przegl Chir. 2012;84:406-10.
30. Chillón Sempere FS, Domínguez Hinarejos C, Serrano Durbá A et al. Quistes epididimarios en la infancia [Epididymal cysts in childhood]. Arch Esp Urol. 2005;58:325-8.