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Case Report

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Cystic dysplasia of the rete testis

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Abstract

Cystic dysplasia of the rete testis is a benign tumor of the testis, which occurs most frequently in pediatric age and is often accompanied by renal anomalies. It is necessary to differentiate this condition from malignant testicular tumors. Different approaches, such as orchiectomy, testis-sparing surgery, or conservative follow-up, can be utilized in treatment. Herein, we reported a five-year-old patient with cystic dysplasia of the rete testis who was admitted with scrotal pain and underwent orchiectomy.

Keywords: Child, cyst, dysplasia, rete testis.

Cystic dysplasia of the rete testis (CDRT) is a rare cause of benign testicular lesions in the pediatric age. It is characterized by irregular cystic dilatations in the rete testis.[1] The condition is often accompanied by renal anomalies, such as ipsilateral renal agenesis or multicystic dysplastic kidney.[2] There is no standardized approach to treatment. Patients treated with orchiectomy or testis-sparing surgery have been reported, as well as those with spontaneous regression during nonoperative follow-up.[2-4] In this case report, we described a patient followed and treated for CDRT.

CASE REPORT

A five-year-old male patient was admitted with complaints of pain and swelling in the left scrotum, which had started two days prior. There was no history of testicular trauma or infection. The patient was being followed with a diagnosis of multicystic dysplasia in the left kidney. Physical examination revealed that both testicles were located intrascrotally, and the left testicle was larger than the right testicle. Palpation did not reveal any mass separate from the left testicle. No tenderness or hyperemia was detected in the left scrotum. On scrotal color Doppler ultrasonography, the right testicle was 16×8 mm, and the left testicle was 26×8 mm. A multicystic lesion with thin septations was detected in the left testicle, and normal testicular parenchyma was not detected. Alpha-fetoprotein, human chorionic gonadotropin, and lactate dehydrogenase values were within their respective reference ranges. The patient was assessed by the oncology council, and exploration was decided since testicular malignancy could not be ruled out. The left testicle was evaluated via a left inguinal incision. It was found to be diffusely enlarged and demonstrated spongious characteristics.

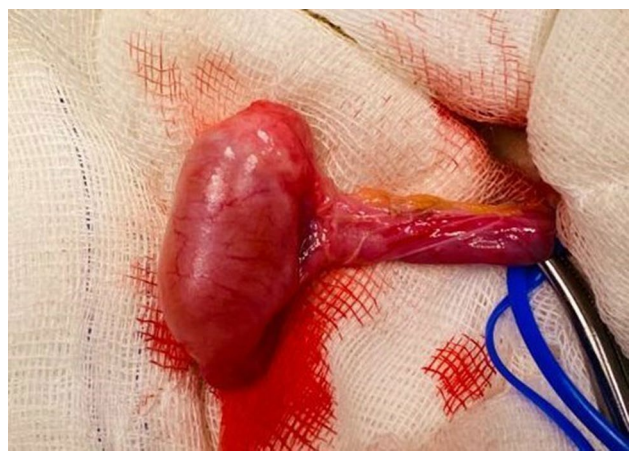

Figure 1. Intraoperative image of the left testicle.

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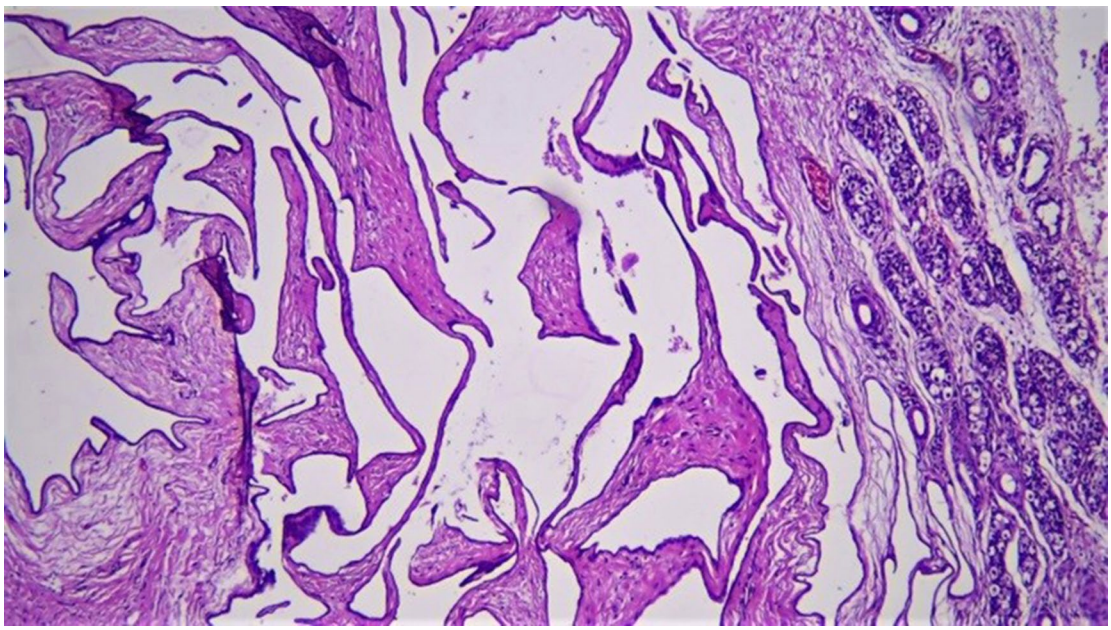

Figure 2. Enlarged cystic structures in the rete testis region, fibrous stroma in between, and immature seminiferous tubules at the edge (H&E, ×40).

Since no mass separate from the testicular tissue was observed or palpated, testis-sparing surgery was not deemed appropriate, and left-sided high-inguinal orchiectomy was performed (Figure 1). Histopathology of the testis revealed cystic dilatation of the seminiferous tubules and multicystic thin septate lesions extending to the tunica albuginea in the rete testis (Figure 2). The findings were reported to be conclusive for CDRT. The patient has since been attending follow-up studies for two years without any issues.

DISCUSSION

Cystic dysplasia of the rete testis is a rare benign lesion and was first described by Leissring and Oppenheimer[4] in 1973. A recent study reported that the number of pediatric patients documented in the English language was 65.[3] The most common presentation in CDRT patients was painless scrotal swelling, and the lesion was unilateral in all reported cases. Pain may rarely develop with the enlargement of the scrotum. The testicle on the affected side may be retractile or undescended. Different from the literature, the patient in our case presented with scrotal pain.[3] The patient also had an accompanying renal anomaly (left multicystic dysplastic kidney).

The pathogenesis of CDRT remains unclear. Leissring and Oppenheimer[4] suggested that CDRT is a degeneration that occurs due to a defect in the connection between the germinal epithelium at the level of the rete testis and the mesonephric duct, causing small cysts in the mediastinum of the testis. They stated that this hypothesis also explained the concomitance of CDRT and renal anomalies. It was reported that progressive ductal dilatation at the rete testis level may cause compression of the normal testicular parenchyma and its displacement with the testicular tissue.[5] Nistal et al.[1] suggested that dilatation occurs as a result of excessive secretion in lumenless immature seminiferous tubules. They attempted to explain the spontaneous regression of cysts with the theory of resorption of fluid after recanalization in seminiferous tubules.

Cystic dysplasia of the rete testis should be differentiated from simple cysts, inflammatory cysts, traumatic cysts, tunica albuginea cysts, and testicular malignant lesions. Scrotal and urinary tract ultrasonography should be performed, and testicular tumor markers should be ordered for differential diagnosis.[4,5] The characteristic sonographic finding of CDRT is the presence of multiple small cysts within compressed normal testicular tissue.[2] Evaluating the testis with magnetic

resonance imaging may provide information on the differentiation of malignant and benign masses.[2] In our patient, scrotal color Doppler ultrasonography revealed a multicystic lesion with fine septations in the left testicle and absence of normal testicular parenchyma. Urinary system ultrasonography showed multicystic dysplastic kidney on the left.

There is no consensus regarding the treatment of CDRT. Contini et al.[3] reported that orchiectomy (48.5%) was performed most frequently with a prediagnosis of malignancy in reported cases. Apart from this approach, testis-sparing surgery (16.7%), nonoperative follow-up (16.7%), perioperative biopsy (4.5%), or testicular exploration without biopsy and excision (3%) have been utilized.[3] Recurrence was reported in four out of 11 patients who underwent testis-sparing surgery.[3] One of these patients was followed up conservatively, two underwent enucleation again, and one underwent orchiectomy. Spontaneous regression (45.4%) or persistence of the cyst (36.3%) was reported in subjects with nonoperative follow-up, and none of these cases were reported to have required surgery during follow-up.[3]

The patient was assessed by the oncology council, and testicular exploration was decided since a possible testicular malignancy could not be ruled out. Although inguinal incision is frequently used in patients undergoing such surgery, the use of the scrotal raphe incision has also been reported.[2] Since the patient was suspected to have malignancy, the testis was explored via a left inguinal incision. Testicular tissue was found to be larger than normal and appeared to be spongious without any palpable mass. Left high-inguinal orchiectomy was performed since testis-sparing surgery could not be performed.

In the literature, it was reported that histopathologic examination of CDRT showed dilatation of the rete testis and seminiferous tubules and compression of the testicular parenchyma; cystic areas were lined with cuboidal epithelium, and the cysts could be microscopic or a few millimeters in size.[4] Histopathological examination of the orchiectomy material of the patient described in this report revealed cystic dilatation of the seminiferous tubules and multicystic thin septate lesions extending to the tunica albuginea in the rete testis.

Since CDRT is a rare anomaly, studies concerning CDRT in the literature are usually at the case report level. Although spontaneous regression during follow-up has been reported, no consensus on treatment has been reached due to the limited number of patients reported in the literature and the lack of information about the development of malignancy in these patients at a later stage. Since our patient did not have normal testicular tissue and a possible malignancy could not be ruled out, left orchiectomy was performed.

In conclusion, CDRT is a rare disease and there is no consensus regarding its treatment. Orchiectomy may be an appropriate treatment option if malignancy cannot be ruled out for the testicular lesion or if testis-sparing surgery cannot be performed due to lack of normal testicular tissue.

Patient Consent for Publication: A written informed consent was obtained from the parent of the patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

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