Case Report

DOI: 10.4274/uob.galenos.2020.1681

Bull Urooncol 2021;20(3):189-191

Unique Approach to Paediatric Sertoliform Cystadenoma of the Rete Testis: A Case of Testis-Sparing Surgery

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Abstract

In this case report, we presented a case of pediatric sertoliform cystadenoma of rete testis which was reported for the second time in the literature and treated for the first time with a testis- sparing approach. A 12 years old boy presented with painless mass in the scrotum. Magnetic resonance imaging showed extratesticular, 4.7x3.8 centimeter, well-circumscribed solid mass with heterogenous high degree vascularity at the postcontrast series, deplasing right testicle to inferomedial. The patient underwent tumor resection without any complication. Histologic examination revealed 5x5x3 centimeter nodular mass containing uniform tubular adenomatous tumoral proliferation. Immunohistochemical examination of the tumor cells showed positivity for panCK, vimentin and WT1. A diagnosis of sertoliform cystadenoma was rendered in the case. The current case is the largest and the second pediatric case in the literature. Sertoliform cystadenoma is a benign extratesticular mass and can be treated with a testis sparing approach.
Keywords: Sertoliform cystadenoma, testis-sparing surgery, pediatric

Introduction

The majority of scrotal masses originate from the testis and most of them are malignant. Paratesticular masses are less common and mostly benign pathologies originating from spermatic cord, epididymis or rete testis (1). They are often indistinguishable from testicular masses due to the close anatomical relationship between the accessory organs and the testis.

Sertoliform cystadenoma is a very rare benign paratesticular neoplasm originating from rete testis. It was first described by Jones and Young (2) in 1997 with a report of two cases. Although it is known to be a benign mass, orchiectomy has been performed in all cases in the literature because of its clinical presentation cannot be differentiated from testicular tumors.

Only 24 cases have been reported in the literature, and only one is pediatric. In this case report, we presented a case of pediatric sertoliform cystadenoma of rete testis which was reported for the second time in the literature and treated for the first time with a testis-sparing approach.

Case Presentation

A 12 years old boy presented with painless mass in the scrotum for one month. Routine biochemical serum and urine test results, beta-human chorionic gonadotropin (0.14 mIU/mL), alpha-fetoprotein (1.91 ng/mL) and lactate dehydrogenase (261 U/L) levels were within the normal ranges. On ultrasonography, both testis were normal, no hydrocel and no focal abnormalities noted in both testicles, pathological retroperitoneal lymphadenopathy had not been detected. But ultrasonography showed extratesticular solid mass at right hemiscrotum that probably orginated from proximal of spermatic cord. Testis parenchyma and margine of the mass were unrelated. The mass had displaced right testicle to inferomedial. Firstly the lesion was considered as benign nature because of the being extratesticular. However, resection of the mass was recommended to the parent of patient because of high degree vascularity of the mass. Magnetic resonance imaging showed extratesticular, 4.7x3.8 centimeter, well-circumscribed solid mass with heterogenous high degree vascularity at the post contrast series, deplasing right testicle to inferomedial (Figure 1). The patient was taken to

Cite this article as: Özman O, Yazıcı G, Özalevli M, Gökmen E, Gönültaş S, Çengel F, Kımıloğlu E, Arslan B. Unique Approach to Paediatric Sertoliform Cystadenoma of the Rete Testis: A Case of Testis-Sparing Surgery. Bull Urooncol 2021;20(3):189-191

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Received: 13.05.2020 **Accepted:** 18.05.2020

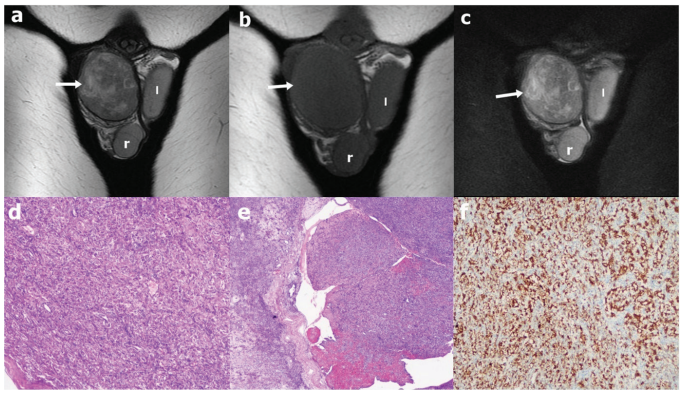

Figure 1. Mass lesion (arrow) with heterogeneous signal intensity, extending from the scrotum into the distal inguinal canal, displacing the right testis (r) inferomedially. T2-weighted without fat suppression coronal magnetic resonance image section (a). Coronal T1-weighted without fat suppression image (a) shows mass (arrow) mainly isointense to the testicular parenchyma (b). Coronal T2-weighted with fat suppression image (c). (l: left testicle). Uniform tubular adenomatous tumoural proliferation (haematoxylin and eosin staining [H&E], ×100) (d). Microscopic cystic space in the focal area (H&E, ×40) (e). Sertoliform cystadenoma with pancytokeratin positivity (pancytokeratin, ×100) (f)

the operation room for right radical orchiectomy after informed consent was obtained from the parents. An inguinal incision was made and the contents of the right scrotum were taken out from the incision with intact tunica vaginalis. The mass, which seemed to be quite separate from the testis, was re-examined perioperatively. After scrotal exploration, the mass was thought to be an extratesticular mass. At this stage, a second interview was conducted with the parents of the patient, and it was stated that the mass could be removed by sparing the testis and informed about the possible risks and benefits. The patient underwent tumor resection without any complication.

Histologic examination revealed 5x5x3 centimeter nodular mass containing uniform tubular adenomatous tumoral proliferation (Figure 1). There was not any mitotic activity, necrosis or nuclear atypia. Immunohistochemical examination of the tumor cells showed positivity for panCK, vimentin and WT1. The cells did not express calretinin, CK5/6, CEA, D240, HBME-1 and inhibin. The vascular cells of tumor mass showed positivity for CD34. A diagnosis of sertoliform cystadenoma was rendered in the case. The patient was alive without evidence of disease recurrence after 4-month follow-up.

Discussion

Rete testis tumors are rare neoplasms. This accessory organ is often the site of local invasion of primary testicular tumors. Cystic and adenomatoid changes are mostly seen in rete testis lined by low columnar epithelium (1). We presented a case of pediatric sertoliform cystatedoma of rete testis that underwent tumor resection with a testis-sparing approach, unlike previous literature. A testis-sparing approach is preferred for histologically confirmed benign tumors, synchronous or metachronous bilateral tumors, incidentally detected non-palpable and small-volume masses, or for tumor in solitary testis and the pediatric cases (3). The importance of testis sparing surgery in paratesticular masses, especially in those patients with normal tumor markers, is emphasized (4). We did not orchiectomy because of negative tumor markers, paratesticular location of the tumor and no infiltrative relationship with the surrounding tissue.

Sertoliform cystadenoma may have nodular, cystic or cystic-nodular macroscopic features. Most of the cases reported in the literature (including single pediatric case) have cystic nature. Nevertheless, our case showed more rare solid macroscopic features. The largest tumor reported in the literature was 4 cm (5). Our case is 5 (radiologically 4.7) cm in diameter and is now the largest case of sertoliform cystadenoma in the literature.

Lahouti et al. (6) emphasized that tumor cells often show calretinin and inhibin positivity, considering their cases and other reports in the literature. However, our case showed a negative immunohistochemical profile for both markers. The case of Mesa et al. (7) was also negative for calretinin and inhibin. Rete testicular adenocarcinoma is a rare malignant tumor with poor prognosis (6). Therefore, differential diagnosis from sertoliform cystadenoma, which is a benign tumor and does not require further oncological treatment and postoperative surveillance, should be made. Malignant tumors can be distinguished from sertoliform cystadenoma with high mitotic activity. No mitotic activity was observed in the current case.

The current case is the largest and the second pediatric case in the literature. Sertoliform cystadenoma is a benign extratesticular mass originating from rete testis and can be treated with a testis sparing approach.

Acknowledgements

Publication: The results of the study were not published in full or in part in form of abstracts.

Contribution: There is not any contributors who may not be listed as authors.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

Ethics

Informed Consent: The patient underwent right radical orchiectomy after informed consent was obtained from the parents.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Concept: O.Ö., M.Ö., E.G., S.G., B.A., Design: O.Ö., E.G., Data Collection or Processing: G.Y., E.K., M.Ö., Analysis or Interpretation: Literature Search: O.Ö., G.Y., Writing: O.Ö., M.Ö., F.Ç.

References

1. Amin MB. Selected other problematic testicular and paratesticular lesions: rete testis neoplasms and pseudotumors, mesothelial lesions and secondary tumors. Mod Pathol 2005;18(Suppl 2):S131-S145.
2. Jones MA, Young RH. Sertoliform rete cystadenoma: a report of two cases. J Urol Pathol 1997;7:47-53.

3. Giannarini G, Mogorovich A, Bardelli I, et al. Testis-sparing surgery for benign and malignant tumors: A critical analysis of the literature. Indian J Urol 2008;24:467-474.
4. Murcia-Pascual FJ, Gracia-Rodríguez R, Vázquez-Rueda F, et al. Testicular and paratesticular tumors in children. Arch Esp Urol 2016;69:691-697. [Article in English, Spanish].
5. Paluru S, Ulbright TM, Amin M, et al. The morphologic spectrum of sertoliform cystadenoma of the rete testis: a series of 15 cases. Am J Surg Pathol 2018;42:141-149.
6. Lahouti AH, Brodherson M, Larish Y, et al. Sertoliform cystadenoma of the rete testis: report of a case and review of the literature. Int J Surg Pathol 2017;25:555-558.
7. Mesa H, Gilles S, Datta MW, et al. Comparative immunomorphology of testicular Sertoli and sertoliform tumors. Hum Pathol 2017;61:181-189.