Journal of Surgical Techniques and Procedures

Research Article
Published: 30 Mar, 2018

Testis-Sparing Surgery for Benign Pathologies in Children: A Report of Two Cases

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Abstract

Aim: The aim of this report is to describe two cases of testis-sparing surgery and to discuss the feasibility, safety and outcome of this treatment modality.

Patients and Methods: We here present the cases of two boys, a 12-year-old and a 7-year-old, exhibiting a Large-Cell Calcifying Sertoli Cell Tumor (LCCSCT) and a Leydig Cell Tumor (LCT) respectively. All operations were performed through an inguinal approach by cold ischemia and with organ-sparing surgery.

Results: Neither of the patients exhibited intraoperative or postoperative complications. After a mean follow-up of 25 months (range 16–34) the patients were free of disease.

Conclusion: Testis-sparing surgery by cold ischemia and frozen-section examination is advisable for pediatric patients with benign testicular masses.

Keywords: Testis-sparing surgery; Children; Leydig cell tumor; Sertoli cell tumor

Introduction

Until the 1980s, orchiectomy was generally considered to be the sole therapy for testicular nodules, particularly with pediatric patients. There is, however, a risk at present of over treatment by radical orchiectomy, especially for small volume lesions detected by Ultra Sound (US) [1]. The high level of accuracy achieved by Frozen-Section Examination (FSE) for identification of both benign and malignant lesions, and the increasing attention paid to the cosmetic, functional and psychological outcomes for young patients with testicular tumors strongly support an organ-sparing approach. The aim of this report is to describe two cases of testis-sparing surgery and to discuss the feasibility, safety and outcome of this treatment modality.

Patients and Methods

Prior to surgery, the two patients had undergone complete staging including clinical examination, scrotal US and CT scans (abdominal and lung). Furthermore, the tumor markers Alpha-Fetoprotein (AFP), beta-human Chorionic Gonadotropin (bhCG) and Lactate Dehydrogenase (LDH) were evaluated to rule out a potential malignancy. Ultrasound was performed using a high-frequency linear- array transducer (8-13 MHz) that allowed for high-resolution imaging of the testicles with a maximum lateral spatial resolution of 0.1 mm.

Case 1

A previously healthy 12-year-old male was admitted to our clinic for pain in their right testis. Sonographic examination of their scrotum revealed a focal 10 mm-sized lesion characterized by a curvilinear acoustic interface with shadowing consistent with a dense area of calcification in the right mid testis. The left testis and the remainder of the right testis were unremarkable. Macroscopically, the tumor was well circumscribed and it had a maximum diameter of approximately 10 mm. The cut surface exhibited a yellow-white solid mass. Histologically, the tumor was composed of large neoplastic cells with abundant eosinophilic cytoplasm with a tubular, trabecular and solid arrangement and loose myxoid stroma with irregularly shaped calcification. Immunohistochemically, the tumor cells were positive for vimentin, S-100 protein, calretinin, inhibin-alpha, melan-A and CD10, while type IV collagen and laminin were observed in the extracellular matrix around the tumor cells [2]. The final diagnosis was a Large Cell Calcifying Sertoli Cell Tumor (LCCSCT) that had been fully excised (Figure 1,2).

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Received Date: *10 Jan 2018*

Accepted Date: *29 Mar 2018*

Published Date: *30 Mar 2018*

Citation:

Marte A, De Rosa L. Testis-Sparing Surgery for Benign Pathologies in Children: A Report of Two Cases. J Surg Tech Proced. 2018; 2(1): 1013.

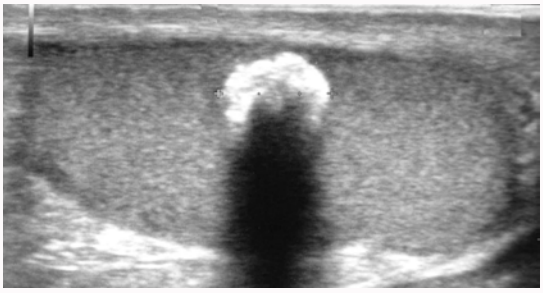

Figure 1: Case 1 Focal 10 mm-sized lesion characterized by a curvilinear acoustic interface with shadowing consistent with a dense area of calcification in the right mid testis.

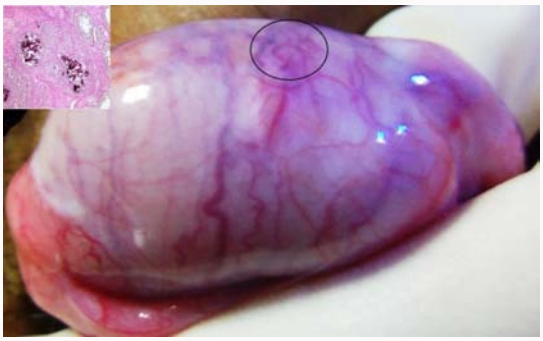

Figure 2: Macroscopic appearance and histology of a Large Cell Calcifying Sertoli Cell Tumor (LCCSCT).

Case 2

A previously healthy 7-year-old male was admitted to our clinic for a small left nodule detected during a testicular US for hydrocele on ultrasound, the testis exhibited a well-defined upper pole focal 6 mm-sized lesion, hypoechoically localized to the upper pole of the testis. The surgery was performed with the same procedure used for Case 1. After delivery of the testis, it exhibited a well-defined yellow nodule at the upper pole that was excised with a wedge resection. The FSE confirmed the presence of a small Leydig cell tumor with unscathed resection margins. The histology revealed medium to large polygonal cells with abundant eosinophilic cytoplasm and distinct cell borders, round nuclei and prominent nucleoli consistent with a Leydig cell tumor that could be fully excised [3] (Figure 3). Both of the operations were performed through an inguinal approach by cold ischemia. The testis and surrounding tunica vaginalis we re-extracted from the scrotum. A soft clamp was applied at the highest level of the cord and the testis and spermatic cord were cooled by surrounding them with a frozen and granular sterile saline solution. The tunica vaginalis was then opened at the closest point to the tumor. Intraoperatively, the location of the tumor was determined by US, or by palpation (when possible). The lesions were isolated by incising the testis with a cuneiform resection of the nodule and the material was sent for FSE. Additionally, biopsies were taken from the surrounding remaining tissue to verify that no solid tumor had been left behind and to confirm testicular intraepithelial neoplasia. Once the pathologist report confirmed the diagnosis, the parenchyma was sutured and the frozen saline was removed along with the clamp. Follow-up consisted of clinical examination, abdominal and scrotal sonography every 3 or 6 months and an annual chest X-ray in LCT patients. Urological follow-up was then scheduled every 6 months for two years, as well as an annual chest X-ray if clinically indicated.

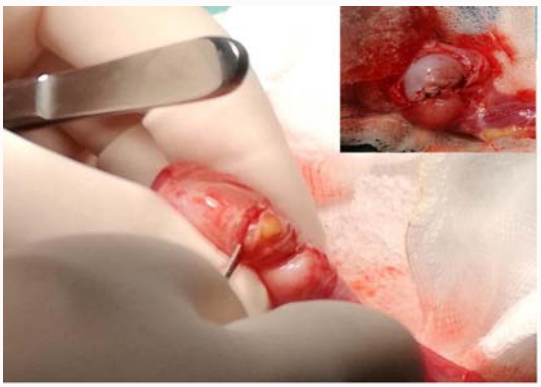

Figure 3: Case 2 Leydig- cell tumor. Excision of the lesion with "Cold ischemia technique" and suture of the testis.

Results

No significant intraoperative bleeding was observed. Neither perioperative infections nor hematomas occurred; no other perioperative or late complications were encountered. Definitive histology did not report any malignant histopathological features in either of the patients. The CT scans did not reveal abnormalities in either of the patients. Both of the patients were free of disease after a mean follow-up of 25 months (range 16-34).

Discussion

Testicular tumors are a rare occurrence in childhood and they are benign in 25% of cases [4]. In light of their uncommon occurrence at a young age, the initial data regarding testis-sparing surgery have been obtained from adult patients. A definite diagnosis should be made by excisional biopsy [5]. None of the other preoperative tests can clarify the exact diagnosis. For this reason, all testicular masses should be assumed to be malignant, until proven otherwise. This is particularly so in pediatric surgery, and informed consent should always include the possibility of an orchidofuniculectomy. There is usually a dissection plane between healthy tissue and the tumor mass. Preoperative and sometimes intraoperative ultrasonography is extremely helpful and enables the surgical team to perform enucleation. Once the frozen-section has confirmed that the tumor is benign and that the surgical borders are tumor-free, vascular occlusion should be terminated immediately and orchiopexy performed. In both of our cases, the results of frozen section and pathological examinations of paraffin blocks were identical. Some authors have described warm ischemia conditions lasting less than 30 min during the occlusion of vascular supply and no testicular atrophy was reported in these cases [6]. This is probably due to relatively shorter periods of ischemia. The development of late testicular atrophy has been reported in an experimental study and some authors strongly recommend cold ischemia conditions [7,8]. In our opinion, cold ischemia is very useful for ensuring a wider safety margin both for the surgeon and for the pathologist, for a possible deepening or to increase the resection margins. Testicular ultrasonography was performed in both of our cases, demonstrating high sensitivity, although MRI can be an efficient diagnostic tool for uncertain cases [9]. LCCSCT is an exceptionally rare neoplasm originating from sperm cord cells. The lesions may occur in an isolated form or in approximately 40% of cases it may be associated with genetic abnormalities, by and large Peutz-Jeghers syndrome and Carney complex. For these reasons, after the diagnosis, a careful evaluation of the patient is necessary to exclude a tumor with

syndromic manifestation [10,11]. Our results with these patients are consistent with the findings in the literature: the lesions were small (<15 mm-in size) and negative for tumor markers [12]. The ideal follow-up interval after conservative treatment of benign testicular tumors is still not clear. Patients with Leydig CT should be followed by medical monitoring, including a clinical examination and US, although the frequency of these medical evaluations and the duration are not well defined. Wegner et al. [13] found one local recurrence after conservative therapy and no progressive disease after reviewing 15 patients with LCTs.

Conclusion

Testis-sparing surgery with potential long-term psychological, cosmetic and functional benefits should be used in pediatric patients with testicular masses in which the healthy testicular tissue appears to be salvageable based on ultrasonography and there are normal levels of tumor markers.

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