Conservative parenchymal surgery in testicular tumors

M.R. Ibarra Rodríguez[1], F.J. Murcia Pascual[1], F. Vázquez Rueda[1,2], M. de Lucio Rodríguez[1], A. Siu Uribe[1], S.D. Ramnarine Sánchez[1], A. Escassi Gil[1], R.M. Paredes Esteban[1]

[1]Pediatric Surgery Clinical Management Unit. Reina Sofía University Hospital. Córdoba (Spain).
[2]Professor at the Faculty of Medicine. University of Cordoba. Maimónides Institute for Biomedical Research of Córdoba (IMIBIC).

ABSTRACT

Objectives. Orchiectomy is the most widely used surgical technique in testicular tumors (TT). However, according to tumor size, tumor markers, and histology, tumorectomy can be considered as the technique of choice, since these tumors are mostly benign. We present our experience with conservative surgery.

Materials and methods. A retrospective study of 21 TT cases in 19 patients under 14 years of age treated in our healthcare facility from 1998 to 2018 was carried out. The following variables were analyzed: age, laterality, histological type, evolution, presence or absence of recurrence, and ultrasound and analytical follow-up. The therapeutic attitude used was reviewed while assessing the possibility of testicular preservation in selected patients.

Results. Conservative surgery was performed in 9 TT cases in 7 patients (2 bilateral cases). Mean age was 6 years (0-13 years). 86% of cases started as an asymptomatic scrotal mass. No significant differences were found in terms of laterality. Tumor markers were negative before and after surgery, except in an infant with high alpha-fetoprotein, which was normalized in the postoperative period. The histological study diagnosed 7 stromal TTs (three Leydig cell stromal TTs, one bilateral Sertoli cell stromal TT, one hamartoma, and one fibroma) and 2 germ cell TTs (bilateral epidermoid cyst). Evolution was favorable in all cases, without clinical or ultrasound recurrence.

Conclusions. Conservative surgery of the testicular parenchyma using tumorectomy can be the first therapeutic option in benign tumors and in selected patients with bilateral tumor, since it allows future hormonal and reproductive function to be preserved.

KEY WORDS: Testicular tumor; Orchiectomy: Conservative surgery.

CIRUGÍA CONSERVADORA DE PARÉNQUIMA EN TUMORES TESTICULARES

RESUMEN

Objetivos. La orquiectomía ha sido la técnica quirúrgica clásicamente más empleada en tumores testiculares (TT). Sin embargo, en función del tamaño del tumor, marcadores tumorales e histología, se puede considerar la tumorectomía como técnica de elección, ya que en su mayoría se trata de tumores benignos. Presentamos nuestra experiencia en cirugía conservadora.

Material y métodos. Estudio retrospectivo de 21 casos de TT en 19 pacientes menores de 14 años, tratados en nuestro centro entre 1998-2018. Analizamos las siguientes variables: edad, lateralidad, tipo histológico, evolución, existencia o no de recidivas, seguimiento ecográfico y analítico. Revisamos la actitud terapéutica empleada, con énfasis en la posibilidad de preservación testicular en pacientes seleccionados.

Resultados. Se realizó cirugía conservadora en nueve casos de TT tratados que correspondían a siete pacientes (dos bilaterales). La edad media de presentación fue de seis años (0-13 años). El 86% de los casos debutaron como masa escrotal asintomática. No existieron diferencias significativas en cuanto a lateralidad. Los marcadores tumorales fueron negativos antes y después de la intervención, salvo en un lactante con alfafetoproteína elevada, normalizada en el posoperatorio. El estudio histológico diagnostica 7TT estromales (tres de células de Leydig y uno bilateral de células de Sertoli, un hamartoma y un fibroma) y 2TT de células germinales (quiste epidermoide bilateral). Evolución favorable en todos ellos, sin recidivas clínicas ni ecográficas.

Conclusiones. La cirugía conservadora del parénquima testicular, mediante tumorectomía, puede ser la primera opción terapéutica en tumores benignos y en pacientes seleccionados con tumores bilaterales, con el objetivo de preservar la función hormonal y reproductora futura.

PALABRAS CLAVE: Tumor testicular; Orquiectomía; Cirugía conservadora.

INTRODUCTION

Testicular tumors represent 1-2% of solid neoplasias in childhood[(1)]. They have a prevalence of 0.5-2 cases per 100,000 individuals under 18 years of age, with incidence having remained relatively stable for the last 30 years[(2,3)].

Corresponding author: Dra. María Rosa Ibarra Rodríguez
E-mail address: rosa_ir90@hotmail.com

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Table I. Summary of clinical cases in our series.

Age	*Histological type*	*Location*	*Clinical signs*	*Tumor markers prior to surgery*	*Ultrasound imaging*	*Associations*	*Recurrence and/or atrophy*	*Tumor markers following surgery*	*Biopsy*
13 months	Fibrous hamartoma	Left	Painless testicular mass	Normal	4 cm hyperechogenic mass	None	No	Normal	No
8 years	Leydig cell tumor	Left	Painless testicular mass	Normal	Mass suggestive of epidermoid cyst or teratoma	Ipsilateral cryptorchidism surgery	No	Normal	No
12 years	Leydig cell tumor	Right	Painless testicular mass	Normal	0.7 cm hypoechoic focal lesion at the testis upper pole	Right herniotomy	No	Normal	No
13 years	Sertoli cell tumor	Bilateral	Painful testicular mass and abdominal pain	Normal	Bilateral testicular calcifications, and 7 mm and 5 mm hyperechogenic lesions	Lentiginosis, gynecomastia, hamartomas in the scalp, Carney syndrome	No	Normal	No
9 months	Fibroma	Left	Painless testicular mass	AFP: 10,84 ng/ml Normal BHCG	12 x 5 mm anechoic lesion	Ipsilateral rete testis cyst	No	Normal	No
2 years	Mature monodermal teratoma	Bilateral	Painless testicular mass	Normal	3.2 mm heterogeneous hyperechogenic mass	Right hydrocele surgery	No	Normal	Pre-operative unilateral biopsy
2 years	Leydig cell tumor	Right	Painless testicular mass, early puberty	Normal	10 x 8 mm intratesticular lesion of heterogeneous echogenicity	McCune-Albright syndrome	No	Normal	No

There are 2 age peaks: between 2 and 4 years of age, and after 15 years of age[4]. According to the literature, these tumors typically have a good prognosis, with a benignity rate of nearly 50%, and a 5-year overall survival rate of 99%[5].

They tend to be little symptomatic. Diagnosis is achieved based on physical exploration, imaging tests (especially ultrasound imaging), and tumor markers[6]. Most patients are asymptomatic at diagnosis. The most widely used markers for diagnosis and follow-up include alpha-fetoprotein (AFP), beta subunit of human chorionic gonadotropin (b-HCG), and lactate dehydrogenase (LDH).

Radical orchiectomy has been typically regarded as the gold-standard treatment of testicular masses across all ages[4]. However, owing to the benign nature of most of these tumors, the treatment approach has been reconsidered, with conservative resection of testicular parenchyma gaining importance[7,8].

The objective of our study was to present our experience in the conservative resection (tumorectomy) of primary testicular tumors in the pediatric population.

MATERIALS AND METHODS

A retrospective study of 21 primary testicular tumor cases in patients under 14 years of age diagnosed at our healthcare facility in the last 20 years (1998-2018) was carried out.

Patients having undergone complete orchiectomy and patients with secondary or metastatic tumors were excluded from the study.

The variables analyzed included age at diagnosis, laterality, histological type, evolution, presence or absence of recurrence, and ultrasound and analytical follow-up (normalization or non-normalization of tumor markers following surgery).

In all cases, the surgical approach was inguinal, exteriorizing the testicle and performing an atraumatic clamp of the spermatic cord to prevent bleeding and tumor dissemination. Tumorectomy was carried out under intra-operative ultrasound control, with lesions being marked using a hypodermal needle for location identification purposes. Age and follow-up data were expressed as a median with range.

RESULTS

Median age was 26 months (9 months-13 years). All patients started with an asymptomatic scrotal mass, except one who had a painful testicular mass associated with abdominal pain. Three tumors were located in the left testicle, two tumors were located in the right testicle, and two tumors were bilateral (Table I).

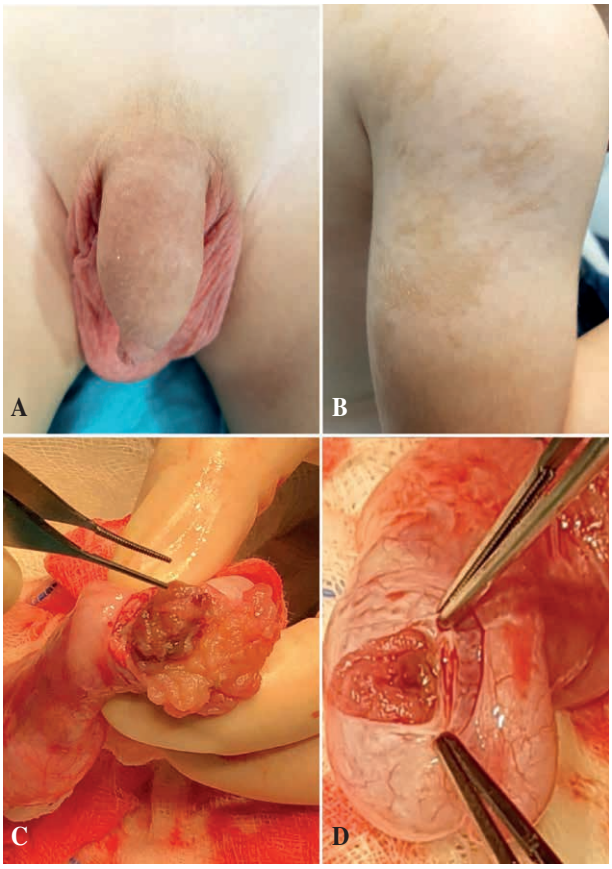

Figure 1. Conservative surgery of a testicular tumor (Leydig cell tumor) in a patient with signs of early puberty and café-au-lait spots, diagnosed with McCune-Albright syndrome. A) Incipient pubarche, a sign of early puberty. B) Café-au-lait spots. C) Intra-operative image. D) Tumorectomy.

Two patients had symptoms typically associated with specific syndromes – one of them had lentiginosis, gynecomastia, hamartomas in the scalp, and frequent fractures compatible with Carney's syndrome, while the other started with early puberty signs and café-au-lait spots, and was subsequently diagnosed with McCune-Albright syndrome (Fig. 1). Other associated pathologies in the series included cryptorchidism, inguinal hernia, hydrocele, and rete testis cyst (all of them ipsilateral).

Following physical exploration, testicular ultrasound examination was performed in all cases, except in the first patient of the series, who additionally underwent magnetic resonance imaging (MRI) after ultrasound examination as diagnosis was unclear. Ultrasound examination demonstrated the presence of an intra-testicular lesion of heterogeneous echogenicity, predominantly hyperechogenic, while MRI showed contrast enhancement in the lesion.

All surgeries demonstrated the presence of well-delimited tumors, with a normal-looking surrounding parenchyma, which was preserved. Following tumorectomy, the remaining of the testicle was reconstructed by suturing the tunica albuginea, and fixated to the tunica vaginalis and the dartos fascia.

In one of the patients, the finding was incidental during the procedure as a result of the presence of a right hydrocele. Following pathological examination, the tumor was diagnosed as a mature monodermal teratoma, with ultrasound examination demonstrating a similar lesion in the contralateral testicle. Bilateral tumorectomy was carried out, with the same pathological diagnosis.

The histological study showed 7 stromal TTs (three Leydig cell stromal TTs, one bilateral Sertoli cell stromal TT, one hamartoma, and one fibroma) and 2 germ cell TTs (a bilateral epidermoid cyst – mature monodermal teratoma).

The tumor markers used were alpha-fetoprotein (AFP) and beta subunit of human chorionic gonadotropin (b-HCG). Both were negative prior to surgery and during subsequent follow-up.

One patient had ipsilateral testicular hypoplasia prior to surgery. None of the patients had post-surgical testicular atrophy. Another patient had bilateral calcifications associated with Sertoli cell tumor.

During follow-up (mean time: 5 years), control ultrasound examinations were performed at month 1, month 3, month 6, and then annually. None of the patients had ultrasound/analytical recurrence, or local/distant metastasis.

DISCUSSION

Testicular tumors are a very rare pathology in the pediatric population[8]. Clinical signs are unremarkable, as demonstrated in our series, with all cases but one presenting as an asymptomatic palpable mass, consistent with findings from other studies[4,7,9].

Various risk factors associated with TT development in all ages have been described: history of cryptorchidism, Klinefelter syndrome, history of testicle cancer in first-degree relatives, and presence of contralateral tumor, among others[4]. In our study, one patient had previously undergone ipsilateral cryptorchidism surgery.

Regarding histology, the literature demonstrates a higher prevalence of germ cell tumors: teratomas, yolk sac tumors, and epidermoid cysts[10,11]. In our series, only 22% of the TTs were germ cell tumors (one patient with bilateral mature monodermal teratoma), similar to Bujons et al., where they represented 33%[9]. Germinal tumors have an incidence of 0.5% in pre-pubertal patients, and 14% in adolescent patients.

Teratoma has a mean presentation age of 13 months (in our series, a single bilateral case was recorded at 24 months of age). Approximately 15% have immature ele-

ments, which are not necessarily associated with a worse prognosis. Tumors of stromal origin are benign in the pediatric population, but malign Sertoli cell tumors have been described in adults. Therefore, **orchiectomy treatment** has been suggested in these patients[9]. Sertoli cell tumors are associated with endocrinological syndromes and disorders (Peutz-Jeghers syndrome, Carney complex, Cushing disease, pituitary adenoma, etc.)[4]. Leydig cell tumors occur at 5-10 years of age (mean age in our series: 7 years), with early puberty. They are usually treated through tumorectomy[13]. In our series, one patient was diagnosed with McCune-Albright syndrome.

According to Taskinen et al., 68% of pre-pubertal patients have a benign tumor, with the proportion of benign tumors being reduced to 38% following puberty[13]. Metastases occur in 61% of adults and only 9% of children. However, in our series, none of the patients had metastasis or died[14].

Tumor markers are useful both for diagnosis and follow-up purposes[15,16]. Indeed, in North American and European protocols, tumor marker negativity is an indispensable criterion to consider conservative surgery[15,17]. AFP levels can be physiologically increased in newborns up to 6 months of age, and also in children with hepatic dysfunction[9]. However, this was not the case in our series.

Although the suspected tumor may be benign considering age and frequency, some groups perform an intra-operative frozen section biopsy to confirm benignity[19]. In our series, biopsy prior to tumorectomy was only carried out in the patient where the tumor finding was incidental.

Testicular atrophy and local recurrence are potential complications of this surgery. Ahmed et al. noted an absence of testicular atrophy in their series following testicular teratoma tumorectomy[20]. In 1984, Weissbach et al. published the cases of two patients with teratoma treated through enucleation, with no evidence of recurrence or testicular atrophy after a 10-year follow-up[10]. In our series, testicular atrophy has not been noted in any patient so far. None of the patients has had local recurrence after a mean follow-up of 5 years.

Until 1980, all testicular tumors were considered malign, so radical orchiectomy was applied in virtually all cases[20]. In the last decades, the therapeutic approach of testicular masses in children has been re-assessed as a result of benignity being predominant in pre-pubertal patients[19,21]. Considering this, as well as how unlikely tumor spread and local recurrence are, tumorectomy looks feasible in this age group, since it allows Leydig cells and seminiferous tubule function to be preserved.

Therefore, although this study has some obvious limitations as a result of being a single-center and retrospective one, we believe conservative treatment through tumorectomy is feasible in patients with a palpable testicular mass and negative tumor markers. However, this procedure should be carried out in an experienced healthcare facility, and the lesion should be completely removed to prevent recurrence.

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