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Benign scrotal masses in children – some new lessons learned

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ABSTRACT

Introduction: A preponderance of benign intratesticular masses in pre-pubertal males encourages testicular-sparing surgery (TSS).
Objective: To review outcome of benign testicular lumps in children managed at a tertiary pediatric center more than 7.5 years.
Methods: A retrospective review of pediatric benign testicular lesions from January 2008 to June 2015 was performed.
Results: There were twelve benign intratesticular tumors. Of these, 11 were in pre-pubertal males; comprising four teratomas, two epidermoid cysts, one dermoid cyst, two cases of Leydig cell hyperplasia, one cystic dysplasia of the rete testis and one large simple intratesticular cyst. We illustrate a case of Leydig cell hyperplasia presenting with precocious puberty limited to the ipsilateral hemi-scrotum. TSS was attempted in all 11 pre-pubertal cases, but successfully performed in seven. TSS was possible for a large testicular cyst seemingly replacing the entire testis, with evidence that the testis reconstituted itself after surgery. Recurrence of an epidermoid cysts reported.
Conclusion: For the first time in the literature, this series reports Leydig cell hyperplasia presenting with ipsilateral hemi-scrotal changes of precocious puberty; shows evidence that the testis reconstitutes itself after TSS for a large cyst; and reports recurrence of an epidermoid cyst after TSS.

Testicular tumors account for 1–2% of all pediatric tumors and have an incidence of between 0.05 and 2 per 100,000 children [1,2]. There are two reported peaks in age; the first is less than 3 years of age and the second in adolescents [1,3]. A high portion of testicular masses in pre-pubertal males are benign [3–5], whereas post-pubertal testicular tumors are mostly malignant [4]. Preponderance of benign disease in pre-pubertal males facilitates consideration of organ preservation. The objective of this study was to review the benign testicular tumors in males at this institution in the last 7.5 years, assess their treatment and outcome, and report on lessons learned that have not been previously published.

1. Methods

A retrospective review was undertaken of all pediatric patients at this institution undergoing surgical excision of intrascrotal masses between January 2008 and June 2015. Cases were identified from both the theater database and the histopathology database. Malignant testicular and paratesticular tumors and tumors in patients with disorders of sex development were excluded from further review. Medical records of cases with benign intratesticular masses were reviewed to determine demographics, clinical presentation, serum tumor markers alpha fetoprotein (AFP) and beta human chorionic gonadotropin (bHCG) and imaging including ultrasound and computer tomography scanning, surgical management, histopathology and outcomes.

2. Results

Twenty-one children underwent surgery for excision of intrascrotal masses, eighteen of whom were pre-pubertal males. Nine of the twenty-one had malignant intrascrotal masses. Seven malignancies occurred in the pre-pubertal age group, four of which were malignant yolk-cell tumors, all with elevated serum AFP. The other three were paratesticular rhabdomyosarcoma. All cases with clinical features of malignancy underwent up-front radical orchidectomy. The two malignancies in the post-pubertal age group included one intratubular germ cell neoplasia in a teratoma and one malignant yolk sac tumor. Both children had markedly elevated serum AFP levels and both underwent radical orchidectomy. Malignant lesions were excluded from further analysis.

There were twelve benign intratesticular lesions in total, eleven of whom were in pre-pubertal males (median age 4 years, range

Abbreviations: TSS, testis sparing surgery; LCH, Leydig cell hyperplasia; LCT, Leydig cell tumor.

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2 months–12.5 years). Hence, eleven of the 18 (61%) testicular tumors in the pre-pubertal group were benign. There were four teratomas (median age 1.5 years, range 5 months–4 years), two epidermoid cysts (age 8 years and 9 years), one dermoid cyst (age 12 years), two cases of Leydig cell hyperplasia (age 3.5 years and 7 years), one cystic dysplasia of the rete testis (aged 4 years) and one large simple testicular cyst (age 13 months). We report for the first time in the literature Leydig cell hyperplasia presenting with signs of precocious puberty limited to the ipsilateral hemiscrotum (Fig. 1a).

In this group of clinically suspected benign intratesticular masses, TSS was attempted in all, and successfully performed in seven cases. Four cases with benign disease proceeded to radical orchidectomy. In two cases with teratoma, the entire testis was replaced by tumor, with no normal testicular tissue visible. One patient with teratoma had a predominance of immature elements on frozen section, raising the suspicion of malignancy. One case had benign Leydig cell hyperplasia, but on frozen section, there was a concern about possible malignancy, which required further staining and analysis to definitively exclude.

The seven patients who successfully underwent TSS are all well and recurrence free at follow-up to date. The intratesticular simple cyst in our series was large and appeared to have replaced the entire testis, bar a thin rim of possible testicular parenchyma (Fig. 2a and b). However, after TSS, we show evidence that the testicular parenchyma stretched thin by the cyst reconstituted itself to a clinically palpable normal testis, with normal parenchymal appearance on USS at one year follow-up (Fig. 2c).

The only benign tumor in the post-pubertal group was one patient with a recurrent epidermoid cyst. The original epidermoid cyst was excised with TSS at age two years at another center, and histopathology at the time confirmed complete excision of a benign epidermoid cyst. The recurrent lesion was noted on an ultrasound done after minor testicular trauma at age 15 years. Repeat TSS was recommended to the patient and family because there was strong clinical suspicion the lesion was likely benign. However, in the context of a strong family history of urogenital malignancies, the patient and family requested a radical orchidectomy after informed consent and review of the case by the hospital ethics committee.

3. Discussion

There is increasing evidence that pre-pubertal testicular tumors are a distinct entity from tumors occurring in the adult testis [3]. Benign testicular tumors occur more commonly than malignant tumors in the pre-pubertal population, and consideration should thus be given to TSS [3,4,6]. Pre-pubertal testicular malignancy treated with orchidectomy has a good prognosis with survival rates of 99% at 5 years [1,6]. Radical orchidectomy for the more common benign tumors results in organ loss that could potentially have been avoided. For this reason it is important to distinguish between benign and malignant tumors pre-operatively.

It may not be easy to clinically distinguish between benign and malignant intrascrotal masses. Most tumors, benign or malignant, present as a painless scrotal mass. There is an associated hydrocoele in 15–50% of cases [1,7]. There are rarely systemic signs to guide diagnosis. Precocious puberty secondary to androgenization may be associated with Leydig cell tumors [7]. The most relevant investigations pre-operatively to guide management are serum tumor markers including alpha fetoprotein (AFP) and beta human chorionic gonadotropin (bHCG), as well as imaging with Doppler ultrasound. AFP and bHCG are important markers of malignancy in testicular tumors. bHCG is produced by syncytiotrophoblast of placenta by tumors including choriocarcinoma and embryonal carcinoma, which are only rarely found in pre-pubertal testicular tumors [7]. However, markedly raised AFP levels are present in 90% of yolk sac tumors which are common in the pre-pubertal population [7]. Benign teratomas may have increased AFP levels, but these are usually not higher than 100 ng/mL [1,3,7–9], so this marker can still be helpful to decide between TSS and radical orchidectomy. On testicular ultrasound, benign tumors tend to be well circumscribed with sharp borders and decreased blood flow [1], and morphology of various lesions such as rete dysplasia (Fig. 3a), simple cysts (Fig. 2a), epidermoids (Fig. 3c) and dermoids (Fig. 3d) can be quite characteristic. Malignant testicular tumors tend to be solid [1]. Malignant tumor metastases are likely to be retroperitoneal or to lung, rarely to the central nervous system or the bone. Twenty percent of yolk sac tumors are associated with lung metastases [1]. There are often no physical signs, so a chest x-ray, computer tomography scan or magnetic resonance scan of the chest, abdomen and pelvis are usually indicated to stage and screen for metastases when a testicular lump is detected [1].

The range of benign pre-pubertal testicular tumors treated at our centre (Table 1) was similar in distribution to other studies in the literature [4]. The current series did not include a case of granulosa cell tumor, also noted in the literature to occur in this age-group. (See Table 2.)

4. Summary of benign lesions

4.1. Teratoma

Four of the 11 benign pre-pubertal cases were testicular teratoma. Teratoma accounts for 45–50% of childhood testicular tumors in studies [4,5,10,11]. It usually presents at the median age of 13 months (range 0–18 months) [1]. Teratomas are mostly benign in pre-pubertal males but often harbor malignancy post-pubertally [12–14]. On ultrasound they often have a heterogenous appearance with areas of calcification [1]. Histology is often pure with diploid DNA content containing all three embryological germ layers, but they can have any combination of the three [1]. In the current study, three of the four teratomas underwent radical orchidectomy, two as a result of the entire testis

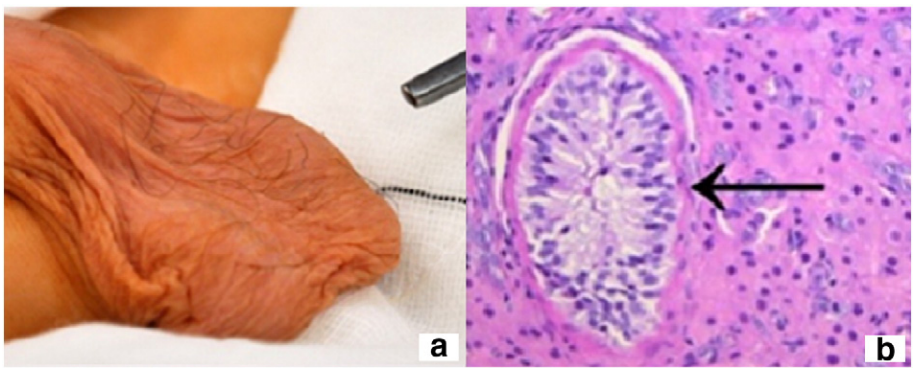

Fig. 1. (a) Leydig cell hyperplasia with ipsilateral scrotal changes of precocious puberty. (b) Histopathology of Leydig cell hyperplasia where Leydig cells are polygonal, with well-defined cytoplasmic borders, abundant eosinophilic cytoplasm and uniform small round nuclei. Entrapped seminiferous tubule (arrow).