

Unifocal Leydig Cell Hyperplasia: Tumor Mimic

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ABSTRACT

Leydig cell tumors and Leydig cell hyperplasia constitute about 3% of testicular lesions in pediatric patients. These lesions are characterized by an interesting constellation of clinical and biochemical features that can facilitate presurgical diagnosis. In some patients, however, these lesions have ambiguous characteristics. We describe here a prepubertal boy with a unifocal Leydig cell hyperplasia that resembled a tumor. The patient was treated successfully by enucleation of the hyperplastic focus.

Keywords: Leydig cell, Tumor, Enucleation.

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INTRODUCTION

Leydig cell tumors present as a testicular mass and are associated with endocrine features that differ according to patient age. In contrast, Leydig cell hyperplasia presents as a diffuse lesion of the testis with no definite masses. We describe here a prepubertal boy with a unifocal Leydig cell hyperplasia that had features resembling a Leydig cell tumor. The patient was treated successfully by enucleation of the hyperplastic focus.

CASE REPORT

Our patient, a boy aged 6 years and 8 months, was brought to our clinic by his parents due to malodorous perspiration. He had no other relevant symptoms or concurrent history of trauma. Upon physical examination, he looked well with no dysmorphic features or acne. He did not have gynecomastia and examination of his genitalia revealed few suprapubic hairs and a nontender, mildly larger right testis with no separately distinguished mass.

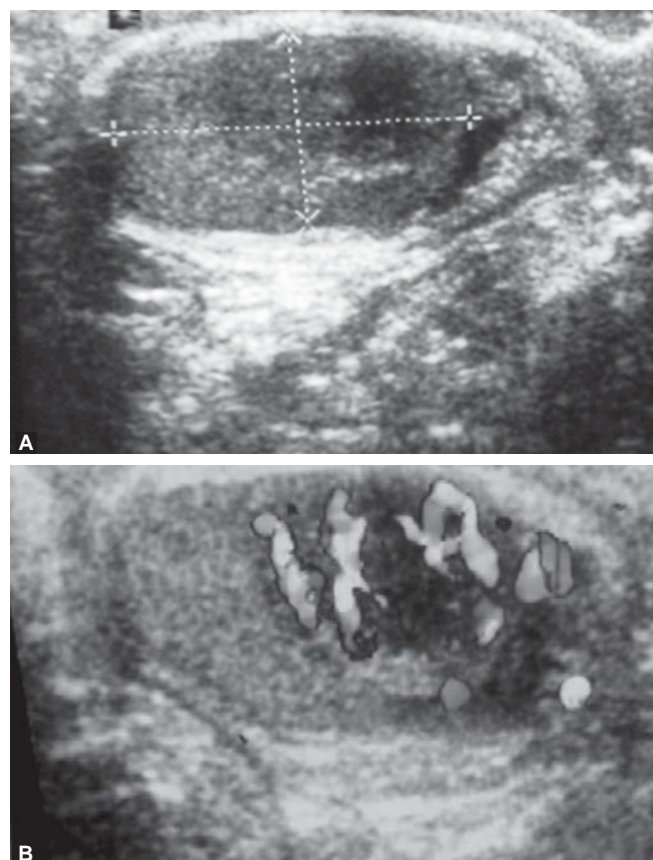
He had a high testosterone (2.72 nmol/L; normal, 0.24-0.69 nmol/l) and 17-OH-progesterone (3.64 nmol/l; normal, 0-3 nmol/l) concentrations. However, estradiol level was within normal range. Scrotal ultrasonography (Figs 1A and B) showed a focal hypoechoic area in his right testis, approximately 13.0 × 6.0 mm in size, with hypervascularity. His serum concentrations of the tumor markers AFP (1.7 IU/ml) and B-HCG (<5 IU/l) were both normal. Bone age from Greulich and Pyle chart was 11 years with a standard deviation of approximately 9 months.

Through an inguinal exploration, clamping of the spermatic cord, the right testis was exteriorized through the wound. The posterior portion of the testis contained a palpable, hardened mass, which was enucleated by blunt dissection after incising the covering tunica albuginea (Figs 2A and B). Light microscopic examination of the mass revealed proliferating Leydig cells wrapped around and entrapping occasional atrophic seminiferous tubules (Figs 3A and B).

DISCUSSION

The incidence of testicular tumors in the general population is low (2/100,000 men)^{2,3} with 3% being Leydig cell tumors (LCT) and hyperplasias (LCH). Of these tumors, 75% affect adults, most often between 25 and 35 years of age, whereas 25% affect children, most often between 5 and 10 years of age.¹

Young, prepubescent patients with Leydig cell lesions frequently show signs of premature virilization with 10%



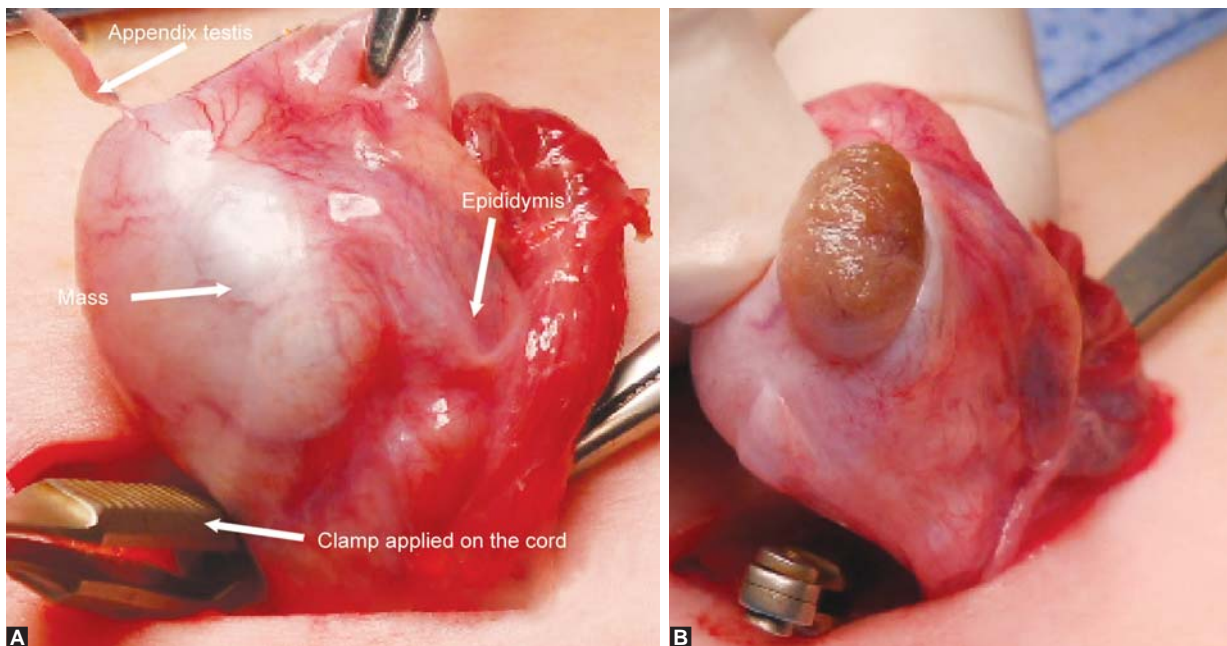
Figs 1A and B: (A) Scrotal ultrasonography showing a focal hypoechoic area in the right testis, approximately 13.0 × 6.0 mm in size, (B) with hypervascularity

presenting with gynecomastia.² Prepubertal boys virilized by Leydig cell tumors have low or normal serum gonadotropin concentrations and high serum concentrations of testosterone, androstenediol, 17-hydroxyprogesterone and 17-ketosteroids.⁴

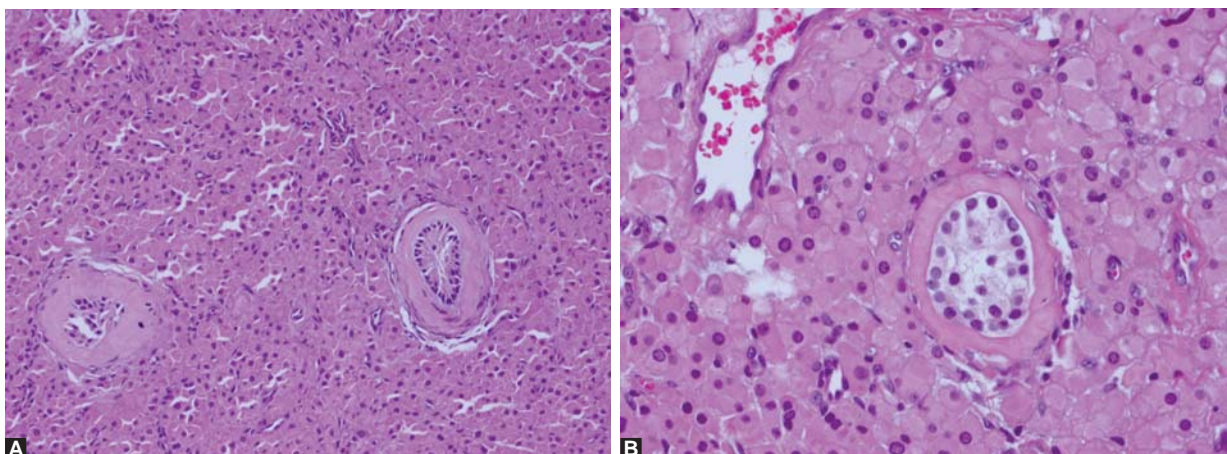
Radiologically, LCT of the testis typically has a homogeneous hypoechoic sonographic appearance (a heterogeneous pattern should alert one to the possibility of a malignancy).⁵ On the other hand, color Doppler imaging, which detects vascular blood flow, often shows hypervascularity of the tumor with a prominent peripheral and circumferential blood flow (Fig. 1B).⁶

Henderson (2006) had believed that these patients are best treated with testis sparing enucleation of the tumor following early occlusion of the spermatic cord, based on the ability to establish the diagnosis preoperatively and the universal benign behavior of unilateral, prepubertal Leydig cell tumor.⁷ However, normalization of testosterone level is expected within 10 days after surgery, while normalization of spermatogenesis and gonadotrophin level occur within several months.¹

Hyperplastic Leydig cells differ morphologically from both normal Leydig cells and Leydig tumor cells. In contrast to Leydig cell tumors, which consist exclusively



Figs 2A and B: Intraoperative pictures: (A) The testis was exteriorized through an inguinal incision with the posterior surface facing anteriorly, with a visible (and palpable) bulging mass. A vascular clamp had already been applied proximal to the spermatic cord, (B) the mass was enucleated through a clear dissection plane after incising the tunica albuginea lying over it



Figs 3A and B: Histologic examination of the testicular mass: (A) Low power view showing sheets of Leydig cells with scattered entrapped seminiferous tubules and small blood vessels, (B) higher power view showing seminiferous tubules entrapped by rather uniform Leydig cells. The latter contained characteristic eosinophilic cytoplasm, small round nuclei and well-defined cell membranes

of Leydig cells, Leydig cell hyperplasia also includes seminiferous tubules containing Sertoli cells. Moreover, Leydig cell tumors are solitary, whereas nodular Leydig cell hyperplasia is characteristically multifocal. In addition, Leydig cell tumors are larger (diameter >0.5 cm) than the foci of Leydig cell hyperplasia (diameter <0.5 cm). However, the size of the lesion was atypical as it was about 1.3 cm in diameter.

CONCLUSION

We have described a prepubertal boy diagnosed with a unifocal Leydig cell hyperplasia that had features resembling a Leydig cell tumor. The patient was treated successfully by enucleation of the hyperplastic focus.

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