



Pediatrics

Cystic Dysplasia of the Testis in an Intraabdominal Undescended Testicle

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ABSTRACT

Cystic dysplasia of the testis is a rare, benign pathologic finding. We present two recent cases of cystic dysplasia of the testis to our practice. The first case involves an 11 month old male presenting with a non-palpable, intra-abdominal left testis. The second case involves a 7 year old male with left scrotal swelling and a history of left renal agenesis. After review of the literature, the first case represents the first report of cystic dysplasia of the testis in an undescended, intra-abdominal testicle.

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Introduction

Cystic dysplasia of the testis (CDT) is a rare, benign testicular tumor presenting primarily in the pediatric population. Scrotal swelling is the most common presentation which prompts referral to the pediatric urologist. Most patients do not report pain with swelling.¹ Commonly these patients will have various other urologic abnormalities such as ipsilateral renal agenesis or multi-cystic dysplastic kidney. We discuss a case of CDT that had a presentation not previously reported.

Case report

The patient was an 11 month old male who presented for outpatient evaluation of a non-palpable left testicle since birth. The patient had no other known physical or anatomical abnormalities. The patient was scheduled to undergo a left laparoscopic orchiopexy for undescended left testis. During the laparoscopic portion of the procedure, an enlarged left testicle was noted at the entrance of the left internal ring. Dissection was carried out in the standard fashion. After placing a 12 mm VersaStep and port through a scrotal incision, the testicle was attempted to be brought down into the scrotum, but would not fit into the 12 mm port. After removal of the

port, the testicle was eventually able to be brought down into the left hemi-scrotum, but would not reach the bottom of the scrotum. Therefore a Fowler–Stevens maneuver was carried out, allowing proper positioning of the testis in the scrotum. Upon further examination of the testis, its size (2.5 cm) and abnormal appearance prompted an intra-testicular biopsy. Upon incision of the tunica albuginea, no normal tubules were seen and appeared to be replaced by a thick fibrous tissue, assumed to be testicular tumor



Figure 1. Picture of the testicle. Caption: Left testicle of patient 1 showing no normal tubular structures, replaced by thick fibrous tissue.

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(Fig. 1). Because of the presumed presence of a testis tumor, a left orchiectomy was performed. Pathologic examination of the specimen revealed cystic dysplasia of the testis with secondary atrophy and fibrosis.

Discussion

CDT was first described by Leissring and Oppenheimer in 1973.² CDT is a rare, benign lesion of the testis, primarily presenting in children at a mean age of 6.1 years.¹ The most common presenting symptom is scrotal swelling followed by cryptorchidism. Scrotal swelling is usually not painful. Prior to this case report it has never been described in an undescended intra-abdominal testicle.

CDT is commonly associated with renal abnormalities. Ipsilateral renal agenesis (51%) and MCDK (21%) are the two most associated renal conditions. Several other urologic abnormalities have been described with CDT including hydronephrosis, cystic epididymis, absent ipsilateral ureteral orifice, ectopic ureter, ureteral duplication, cross renal ectopia, and urethral stricture.¹

Microscopically the cysts are composed of flat or cuboidal epithelial cells separated by fibrous septa.³ The disorder is thought to develop embryologically from failure of the mesonephric duct.² A disruption occurs in the fusion of the mesonephric duct and germinal epithelium at the level of the rete testes. Subsequently a cystic dilation develops in the rete testis. Another theory of pathogenesis consists of abnormal fluid secretion by pre-pubertal rete testis cells.⁴ The fluid cannot escape into the non-mature seminiferous tubules and cystic dilation ensues. Two specific cases of spontaneous regression of CDT around the ages of puberty are

thought to support this theory as the seminiferous tubules form a lumen the fluid can escape, resolving the cysts.^{1,5}

The treatment of CDT is variable and currently there are no guidelines or consensus, thus it tends to be based on the comfort of the urologist and concern for malignancy. The decision of orchiectomy, enucleation or watchful waiting is centered on the desire to preserve endocrine function and fertility. Of those cases reported in the literature, 65% underwent an orchiectomy while 27% underwent a more conservative management of either enucleation or observation.¹ Observation is becoming a more popular option, since this is a benign condition and there have been reports of spontaneous regression.⁵ The degree of involvement of the testis, possibility of lesion recurrence as well as the fear of an underlying malignancy and misdiagnosis given the fact that these are often found incidentally on exploration, continues to drive the treatment of CDT.

Conflict of interest

No conflict of interest.

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