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Adenomatoid tumor of the testis in a child

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Abstract Adenomatoid tumors are rare benign neoplasms thought to be of mesothelial origin. Although most reported cases arise from the epididymis, rare cases have been reported in the spermatic cord, testicular tunica, ejaculatory ducts, prostate, and suprarenal recess. We describe a 4.5-year-old boy who presented with a relatively asymptomatic right testicular mass that was resected and confirmed to be adenomatoid tumor of the testis by histopathology. Because of its rarity, the clinical and histopathologic aspects are discussed.

According to the literature, testicular tumors are rare with an incidence of 0.5 to 2 per 100,000 children [1,2]. The most common histologic features for prepubertal tumors are pure yolk sac tumor and teratoma. Adenomatoid tumors are relatively uncommon benign tumors of mesothelial origin, usually occurring in the male and female genital tracts. Extragenital adenomatoid tumors are rare and have been found in the adrenal glands, heart, mesentery, lymph nodes, and pleura [3]. Recently, we encountered a case of intratesticular adenomatoid tumor in a 4.5-year-old boy. To the best of our knowledge, this is the youngest patient reported with adenomatoid tumor of testis. Herein, we describe this rare case and the associated histopathology and management.

1. Case report

A 4.5-year-old boy presented with a gradual increase in size of the right side of the scrotum of more than 1-year duration unassociated with pain or other symptoms. He had a left undescended testis at birth, which descended to the scrotum by second year of life. His physical examination was unremarkable, except for an approximately 2.0 cm nontender intratesticular mass. Scrotal ultrasonography revealed a 2.6 × 1.6 × 1.4–cm hypoechoic multilocular cystic mass of the right testis, which was presumed to be a cystic teratoma (Fig. 1). However, serum tumor markers including alpha-fetoprotein and beta-human chorionic gonadotropin were within normal limits. The child subsequently underwent testis-sparing surgery because the intraoperative frozen section analysis of the mass showed no evidence of malignancy. Postoperative histopathologic examination of the mass demonstrated irregularly arranged tubules and glandlike structures (Fig. 2A). Diagnostic confirmation was achieved with immunohistochemical tests, which were positive for cytokeratin (Fig. 2B) and vimentin (Fig. 2C) representative of adenomatoid tumor. There was no history of testicular tumor or any other tumor in the family. At the 12 months of follow-up examination, the patient was asymptomatic, and scrotal ultrasound scan showed no evidence of local recurrence. The size of the residual right testis was 1.1 × 0.9 × 0.4 cm, and left testis, 1.7 × 1.1 × 0.9 cm.

2. Discussion

Pediatric testicular tumors are rare [1,4]. Yolk sac tumors and teratomas are most common in prepubertal children.

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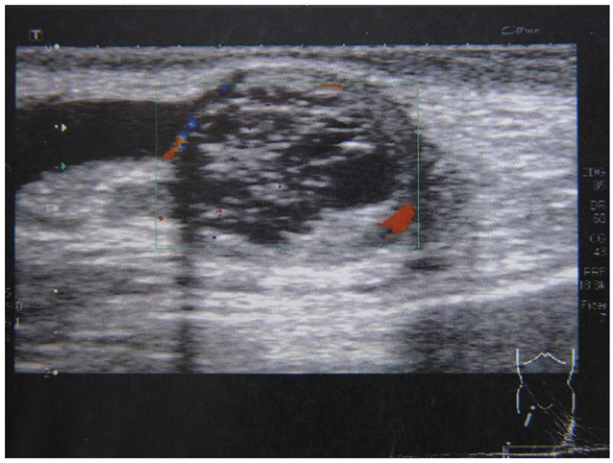

Fig. 1 Scrotal ultrasound scan revealing a 2.6 × 1.6 × 1.4–cm hyperechoic multilocular cystic mass of the right testis.

After the onset of puberty, the most common tumor is embryonal carcinoma. Traditionally, most of pediatric testicular tumors have been considered malignant. Recently, it has been recognized that cystic testicular lesions in children are usually benign [5]. We reported this case because of the rarity of benign intratesticular adenomatoid tumors occurring in children. To date, only a few cases of primary intratesticular adenomatoid tumor have been described with Hostman et al [6] being the first to publish a case describing this condition in children in 1992. To the best of our knowledge, this is the youngest reported child with adenomatoid tumor of the testis.

The term *adenomatoid tumor* was introduced by Golden and Ash in 1945 to describe a group of benign tumors with a glandular pattern and obscure histogenesis, localized in the urogenital tract. Adenomatoid tumors are the most common paratesticular neoplasms and account for approximately 30% of all paratesticular masses [7]. Most adenomatoid tumors are asymptomatic and present as small, solid, nontender masses. The histogenesis of adenomatoid tumors has been a source of controversy. Support for a mesothelial origin for adenomatoid tumor has been derived from ultrastructural studies of both male and female genital tract tumors [8-11]. In our case, the structural growth pattern of adenomatoid tumor was atypical of benign neoplasms because it was not encapsulated and tumor elements were present between the structures of adjacent tissues. The microscopic features of the resected tumor consisted of an unencapsulated mass of neoplastic cells arranged in tubules and glandlike structures. Mitotic figures were not seen. As illustrated in our case and supported by other studies involving adenomatoid tumors from both male and female genital tracts, the tumor exhibited positive immunostaining for cytokeratin and vimentin [7,12,13].

Orchiectomy has traditionally been the gold standard for the treatment of testicular tumors in children. According to recent favorable reports, testis-sparing surgery has become a serious option in the therapy of benign testicular tumors [14,15]. Even in children in whom the testis appears completely replaced by tumor on ultrasonography, enucleation can leave significant normal testicular tissue that was merely compressed by the benign lesion [16]. Frozen section histology has been documented to be a reliable tool for discriminating between benign and malignant tumors [17]. It can thus be used whenever testis-sparing surgery is considered. In this case (serum tumor marker levels within normal limits and no signs or symptoms of metastatic spread), enucleation of the tumor with frozen section histology appears to be the treatment of choice. After the benign histologic type was confirmed, the testicular defect was closed with absorbable suture, and the testis returned to the scrotum.

Adenomatoid tumor is a benign lesion with clinical signs similar to those of other testicular neoplasms. The ultrasonography findings are nonspecific, with the mass presenting with isoechoic, hypoechoic, or hyperechoic nodules. Normal levels of preoperative serum tumor markers combined with intraoperative histologic examination can prevent needless orchiectomy, thereby maintaining endogenous testosterone production and fertility potential at its fullest [18,19]. Progress is favorable in all cases, with no instances of relapse published. Follow-up by postoperative ultrasound is sufficient.

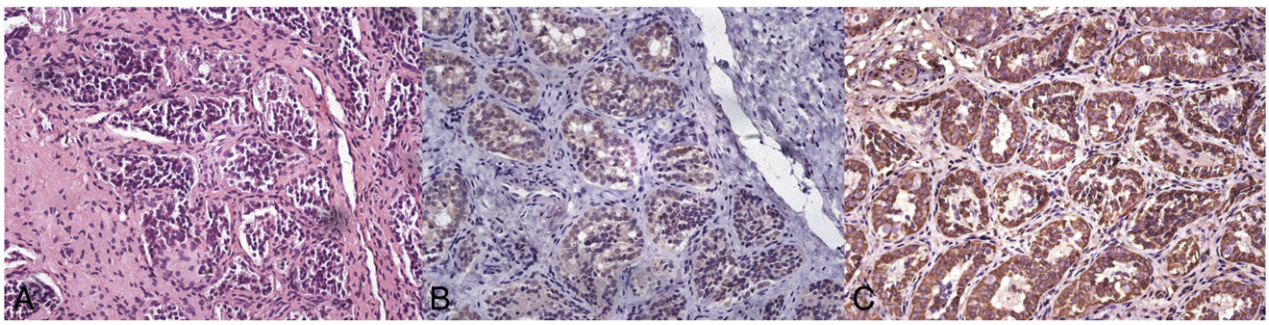

Fig. 2 Photomicrograph of hematoxylin-eosin stain of testicular biopsy specimen in A. Cytokeratin (B) and vimentin (C) labeling are localized within the cytoplasm of tumor cells (original magnification ×200).

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