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Adenomatoid paratesticular tumour – Rare presentation in a child

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Introduction

Adenomatoid tumours are rare mesothelial neoplasms described, in the urogenital tract, in 1945 by Golden and Ash.[1] Accounting for approximately 30% of all paratesticular masses, they are the most common paratesticular neoplasms.[1,2] In males, they can occur in the epididymis, the testicular tunics, the spermatic cord and intratesticular.[2] These tumours are benign, with minimum infiltrative behaviour, therefore testicle-sparing excision is the treatment of choice.[1,2] We present a rare case of a paratesticular adenomatoid tumour, originating from the tunica albuginea, presenting as an incidental finding of in a fourteen-year-old male. We make a brief review of the physiopathology, clinical presentation and management.

Case presentation

A fourteen-year-old male, with previous history of a surgically corrected proximal hypospadias. In the setting of an investigation for delayed onset puberty, a high left testicle was identified. On scrotal ultrasound, an inguinal left testicule was recognized, with normal morphology but smaller than the contralateral (18mm vs 21mm) and with small venous ectasias, the right testicle was normal. Pre-pubertal endocrinal profile was normal. He was proposed for a left orchidopexy. On inguinal exploration, we found a small left testicule with a yellow-brown coloration, with epididymis-testicular dissociation and a homolateral varicocele (Fig. 1). No mass could be individualized from the testicule macroscopically therefore, to avoid an unnecessary orchidectomy in a patient with delayed puberty onset, a testicular biopsy was made before the orchidopexy was performed. Histopathological analysis from frozen section showed a normal testicule and a paratesticular lesion suggestive of a benign adenomatoid tumour, with fibrous stroma, small cells with round nuclei, without atypia or mitosis, with vacuolated cytoplasm (Fig. 2). Immunohistochemistry tests were positive for cytokeratin AE1/AE3, calretinin (Fig. 3) and beta-catenin and negative for CD34, S100, desmin, CD68 and CD45. Post-operative tumour markers were negative. In the setting of microscopic disease without an identifiable macroscopic mass, excluding the possibility of a testicle sparing surgery, a conservative approach was chosen. He remains under Pediatric Urology surveillance without identifiable paratesticular mass on physical examination and ultrasound, at one-year of follow-up.

Discussion

Pediatric testicule tumours are rare, with an incidence of 0.5–2 per 100 000 children.[1] Yolk sac tumours and teratomas are the most common in prepubertal children and, embryonal carcinoma the most frequent in post pubertal.[1] Paratesticular tumours are less than 10% of intrascrotal neoplasms and mostly benign (70–80%),[2] being adenomatoid tumours the most common. Adenomatoid tumours are normally located in genital tracts, but extragenital locations have been reported in the heart, pleura, mesentery, adrenal glands and in the lymph nodes.[1,2] In males, the most common location is in the epididymis (77%), with rare case reports in the tunica albuginea (14%, as presented in our patient), the testicular parenchyma (7.5%) and the spermatic cord (0.5%).[3]

There are many theories about their histogenesis: mesothelial, Müllerian, mesonephric and endothelial origin, but the most accepted is the mesothelial origin, supported by electron microscopy and immunohistochemistry findings.[2,4]

They present, typically between the third and fifth decades, as a small (2–5cm), solid, painless mass, discovered incidentally or on

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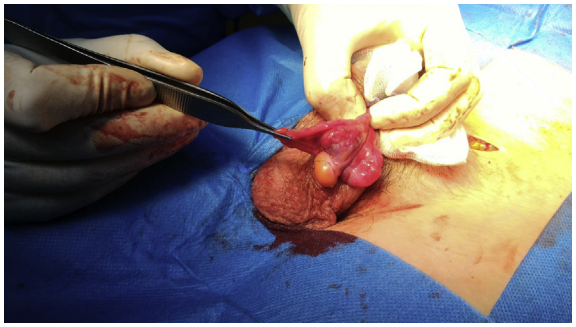

Fig. 1. Intraoperative findings of a yellow, macroscopic homogenous, left testicule. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

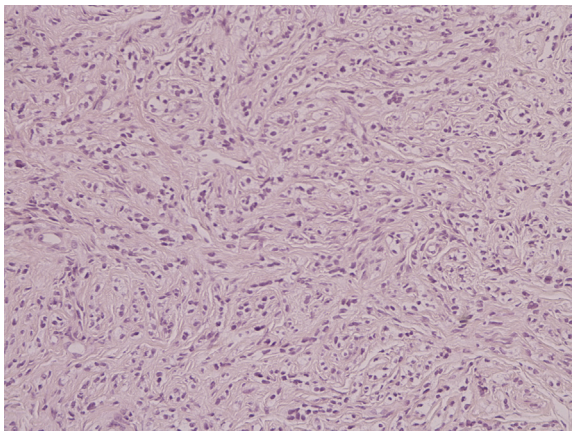

Fig. 2. Proliferation of cords and tubules of cells with vacuolated cytoplasm and small benign appearing nuclei in a fibrous stroma.

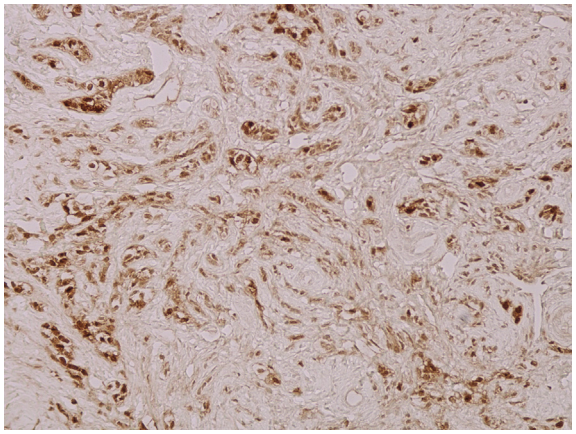

Fig. 3. The proliferating cells strongly expressed calretinin (by immunoperoxidase).

physical examination.[1–3] Rarely, pain can be the presenting symptom.[3] They are predominately located on the left side (56%)[2,3] and up to 20% have an associated hydrocele.[3] Our case report is, therefore, a rare precocious incidental diagnostic, one decade earlier than normal presentation.

Scrotal ultrasound is the preoperative imaging method of choice[4] but the findings are nonspecific.[1] It can distinguish testicular from paratesticular and cystic from solid lesions[4] but can present as isoechoic, hypoechoic or hyperechoic (most commonly the latter).[1,5] Adenomatoid tumours of the tunic are typically benign but can invade the testicule, making it difficult to differentiate from intratesticular tumours.[4] In the case reported no suggestive signs were found on physical examination or on ultrasound.

Preoperative serum tumour markers (alfa-fetoprotein and human chorionic gonadotrophin) are characteristically negative.[4] Hormonal associations have been claimed but are rare and their significance is unclear.[4]

Macroscopically, during surgery, it presents as a small, round, solid mass with elastic consistency and white, yellow or brown coloration.[2–4] A single tumour is most common but multiple small tumours may be seen, especially when involving the testicular tunic.[3] Surgeons should be alert to malignant paratesticular masses, namely a rhabdomyosarcoma, who normally present as large, painless, palpable masses (> 5cm) and require aggressive surgical resection. Macroscopic analysis might be misleading and definitive differential diagnosis can only be made by histology on frozen section.

Microscopically, adenomatoid tumours present as unencapsulated masses with neoplastic cells arranged in three possible patterns: plexiform, glandular (or tubular) and angiomatoid (or canalicular).[1,2] Nuclei are usually spherical and central, and the cytoplasm typically presents vacuoles.[2] No mitotic figures are found.[1] They have positive staining on immunohistochemistry tests for cytokeratin AE1/AE3 and vimentin[1,2] but not for factors VIII and CD34.[2] Their mesothelial origin is confirmed by positive staining for calretinin.[2] In the case presented, no mass was macroscopically identifiable and the diagnose was only made on microscopy with immunohistochemistry.

Adenomatoid tumours have benign behaviour.[2] Nonetheless, nuclear atypia and local invasion have been seen, particularly in epididymal head tumours,[2] as well as local recurrences and one case of possible malignant transformation.[5]

Treatment consists of surgical removal and when feasible, simple enucleation is the choice.[1–3] However, a correct preoperative diagnosis is difficult, leading frequently to unnecessary orchiectomy.[3] The prognosis is good and follow-up by postoperative ultrasound is generally sufficient.[1] In the presence of microscopic disease in an adolescent with delayed onset puberty, were excision could only be made through orchiectomy, the authors present a case of conservative treatment with management though clinical and ultrasound surveillance.

Conclusion

Adenomatoid tumours are rare benign neoplasms with frequent scrotal location. Being difficult to distinguish, preoperatively, from malignant tumours, high suspicion is fundamental to avoid unnecessary orchiectomies. A testis-sparing approach in a child is very important since it preserves hormonal function and has a good cosmetic result, with positive psychological impact. When a testicle-sparing approach is impossible due to absence of macroscopic disease, a conservative approach may be chosen, with regular clinical and ultrasound surveillance.

Appendix A. Supplementary data

Supplementary data related to this article can be found at https://doi.org/10.1016/j.eucr.2018.08.008.

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