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Case Report

Prepubertal-type teratoma in a postpubertal patient: case report and review of literature

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Abstract: Prepubertal-type teratomas are rare, especially in postpubertal patients. We present a case of a 43-year-old man with a palpable painless mass in the left-sided testis discovered by accident. Scrotal ultrasound and magnetic resonance imaging revealed a 2.7×2.0 cm mass inside the left testis, and malignancy could not be excluded, though classical serum tumor makers were within normal limits. Radical testicular resection was then conducted, and the pathologic report proved the mass to be a testicular epidermoid cyst, a rare form of prepubertal-type teratoma. Relevant published literature is also reviewed in our text.

Keywords: Prepubertal-type teratoma, epidermoid cyst, testicular neoplasms

Introduction

According to the 2016 WHO classification of tumors of the urinary system and male genital organs, prepubertal-type teratoma is a newly-defined subtype of germ cell tumor and is considered to be unrelated to germ cell neoplasia in situ (GCNIS) and distinct from postpubertal-type teratoma. Based on our systematic review, prepubertal-type teratomas were rarely referred to in previous studies, especially cases that occurred in postpubertal populations, and few studies described the clinical features, diagnosis, and treatment of prepubertal-type teratoma. Here, we present a rare case of prepubertal-type teratoma occurring in an adult patient, and the existing related literature is reviewed.

Case presentation

A 43-year-old man was admitted to our hospital for a mass in the left testis identified accidentally by sonography during a nonroutine check-up. The physical examination confirmed a palpable painless mass inside the left testis. Further general tests found no enlargement of lymph nodes in the inguinal, retroperitoneal, or supraclavicular regions. No positive clinical symptoms were mentioned by the patient, and the classical serum tumor makers (α-fetoprotein, β-human chorionic gonadotropin, and lactic dehydrogenase) were within normal limits.

The scrotal ultrasound manifested an approximately 2.7×2.0 cm well-demarcated intratesticular mass with alternating echogenicity and a sporadic blood flow signal at its rim (**Figure 1A**, **1B**). Further magnetic resonance imaging (MRI) revealed that the lesion was nonhomogeneous and well-demarcated, and mild enhancement was observed on contrast-enhanced imaging (**Figure 1C**, **1D**). Chest and abdominopelvic enhanced computed tomography were conducted, and neither lymphatic metastasis nor visceral metastasis was detected. Surgical operation was then performed. Frozen sections of intraoperative samples showed elements of teratoma, and a specific pathologic type could not be confirmed. Considering that little normal testicular tissue remained and the lesion had suspected malignant potential, and that the patient was concerned, inguinal radical testicular resection was carried out.

Grossly, the lesion was sharply demarcated with a yellow amorphic mass inside (**Figure 2A**). Microscopically, a squamous epithelium-lined cystic cavity containing yellow to gray kerati-

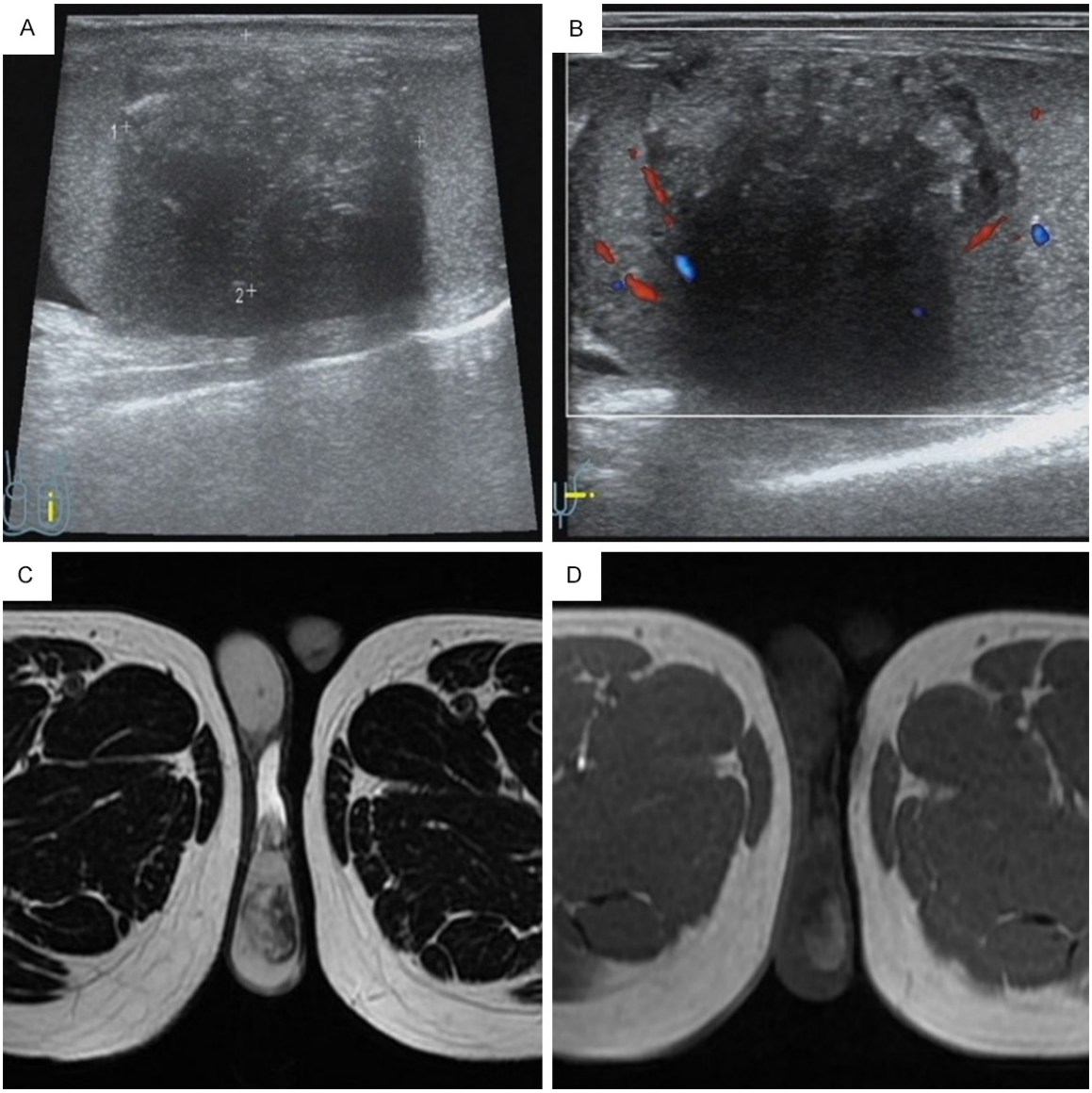

Figure 1. A. 2.7×2.0 cm well-demarcated intratesticular mass with alternating echogenicity. B. Sporadic blood flow signal at the rim of the mass. C. Nonhomogeneous well-demarcated lesion inside the left testis. D. Mild enhancement on contrast-enhanced MRI.

nous material was observed, and no cutaneous adnexal structures were discovered (**Figure 2B**). At high power, spermatogenesis around the mass was observed, hyaline degeneration was noted in some parts of the seminiferous tubules, and no GCNIS was detected (**Figure 2C**, **2D**). Overall, the mass was histopathologically verified to be an epidermoid cyst.

Discussion

Testicular epidermoid cysts (TECs) were first described by Dockerty and Priestly at the Mayo Clinic in 1942 [1]. In the recent WHO classification of 2016, TECs are listed as teratomas of the prepubertal type within the group of germ cell tumors unrelated to GCNIS. Previously, teratomas in the postpubertal testis were considered to be malignant, and in recent decades, there has been increasing evidence supporting the existence of benign teratomas in postpubertal patients [1, 2]. Nonetheless, prepubertal-type teratomas in postpubertal men are still very uncommon. In most situations, epidermoid cysts occur in the skin of the scalp, ear, face, and back. It has been reported that TECs

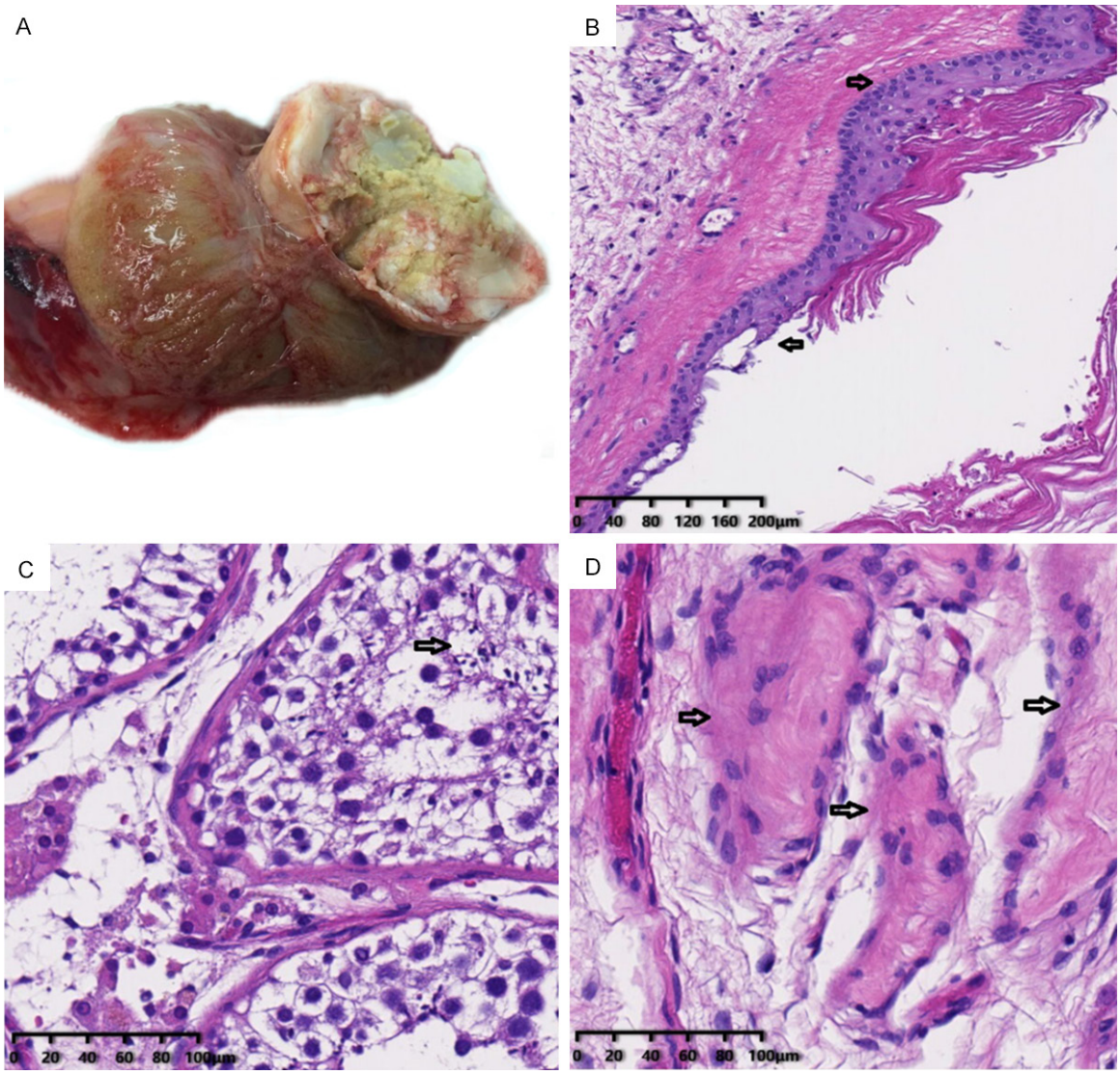

Figure 2. A. Sharply demarcated cut surface with a yellow amorphic mass inside. B. Squamous epithelium-lined cystic cavity without cutaneous adnexal structures (H&E 1:200). C. Spermatogenesis around the mass, which was not GCNIS (H&E 1:400). D. Hyaline degeneration in part of the seminiferous tubules (H&E 1:400).

constitute only 1% of all testicular neoplasms [3].

Typically, TECs present as a palpable painless intratesticular mass incidentally discovered during self-examination or routine physical examination, usually detected in the second and fourth decades of life, with the right side being the predominant site of lesions [1, 4]. Occasionally, patients may present with symptoms of scrotal pain or vague discomfort, similar to those of malignant testicular tumors. The pathogenesis of TECs remains controversial and incompletely understood, and a monodermal development of a teratoma or squamous metaplasia of the rete testis or seminiferous epithelium may be a reasonable interpretation [3, 5]. Serum tumor makers are usually in the normal range, and no metastases have been reported [1, 5].

TECs share unique ultrasonographic characteristics, mainly presenting in the form of a well-demarcated roundish-shaped lesion with a hyperechogenic rim, no or little intralesional echogenicity, and acoustic shadowing on the posterior edge of the tumor [1, 6]. The classical "onion-ring" appearance with alternating hyper-

echoic and hypoechoic layers was first described by Malvica in 1993 and is present in approximately 60% of all cases, which corresponds to the alternating layers of keratinizing squamous epithelium, compacted keratin, and desquamated epithelium seen pathologically [6]. Contrast-enhanced ultrasound demonstrates the absence of contrast bubbles within the lesion, indicating an avascular area, and real-time elastography depicts that epidermoid cysts are hard, which will help distinguish them from other similar lesions. It should be noted that an atypical sonographic appearance has been reported [7], in addition, the hyperechoic nodule with marked acoustic shadowing and a surrounding hypoechoic area, which suggest a calcified tumor, make TECs difficult to distinguish from a burned-out tumor [1, 5]. On the other hand, the "onion-ring" appearance can also be seen in other types of teratomas [4, 6]. Scrotal MRI can likewise show the abovementioned morphologic features, commonly presenting as sharply demarcated "bulls-eye" intratesticular lesions with increased signal intensity at the rim of the lesions and no enhancement of the central area, and T2-weighted imaging shows high intralesional signal intensity [1, 6].

Histopathologically, TECs are characterized as intraparenchymal squamous epithelium-lined cystic cavities filled with keratinized material without cutaneous adnexal structures; have an organoid architecture; lack significant cytological atypia, GCNIS, tubular atrophy and scarring, impaired spermatogenesis, and microlithiasis; and largely lack chromosome 12p amplification (except in a recently debated study) [8, 9]. Ciliated respiratory epithelium as well as well-developed and thick bundles of smooth muscle have been reported to be frequent, sometimes creating an organoid bronchus-like structure; by contrast, intestinal differentiation has been relatively uncommonly reported [8]. Gross findings include a cystic structure filled with yellow to gray keratinaceous debris [5].

Epidermoid cysts are generally thought to be nonmalignant, and the management of TECs tends to be a more conservative approach. Organ-sparing surgery has been performed successfully in several reports, but a standardized diagnostic pathway is required, and frozen section examination may be utilized for determining the need for surgery [10-12]. It was suggested that benign testicular cysts may affect the normal parenchyma of the testis even if the histopathology is normal. Thus, the excision of these benign lesions, especially in the infertile population, is recommended [13]. TECs are generally deemed to be biologically benign, and an excellent prognosis is anticipated [5, 12]. However, malignant transformation to basal cell carcinoma or squamous cell carcinoma from epidermoid cysts has been reported, and careful observation is recommended [12, 14].

Conclusion

A prepubertal-type teratoma, namely, a testicular epidermoid cyst, in a postpubertal patient is rare, and the common clinical manifestations as well as the typical characteristics on radiographic and histopathologic examinations should be unambiguously recognized. Testis-sparing surgery can be performed if the tumor volume is small relative to the entire testis, and a good prognosis can be expected, although close follow-up is needed.

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Disclosure of conflict of interest

None.

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