

Epidermoid cyst of the testis: a report of six cases

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Abstract

Introduction: Testicular epidermoid cysts are rare tumors that occur in 1% of testicular tumors and are often misdiagnosed as malignant lesions clinically. We report six cases of epidermoid cysts. **Case Presentation:** The chief complaints were scrotal induration in five patients and pruritus scrotum in one patient, and the patients were aged

Case report

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Key Clinical Message

Testicular tumor markers are negative in patients with epidermoid cysts. Clear margins and sole and small testicular tumors (25 mm or less) suggest the possibility of epidermoid cyst.

Keywords: epidermoid cyst, testicular tumor, ultrasonography

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

Introduction

Testicular epidermoid cysts are rare tumors that occur in approximately 1% of testicular tumors and are often found in young men 10–39 years of age.^{1–4} They are benign tumors that are often misdiagnosed as malignant lesions in clinical practice. Although a preoperative diagnosis can avoid unnecessary orchiectomy, it is often difficult to achieve. Therefore, high inguinal orchiectomy is performed in >80% of the reported cases.¹ Notifying the possibility of a benign tumor (1%) to patients is crucial before surgery. We summarized specific preoperative findings differentiating epidermoid cysts from malignant lesions. Herein, we report six rare cases of epidermoid cysts in the testes.

Case presentation

We encountered six patients with epidermoid cysts at our hospital between April 2010 and March 2022 (Table 1). The median age (range) was 20 (9–30) years. The tumors occurred in the right testes in two patients and the left in the other four patients. The chief complaints were scrotal induration in five patients and pruritus in the scrotum in one patient. The patients had no family or medical history of testicular cancer. Their blood counts, biochemical test results, and levels of testicular tumor markers (lactate dehydrogenase, alpha-fetoprotein, human chorionic gonadotropin, and human chorionic gonadotropin- β) were within the normal ranges. Ultrasonography (US) was performed in all patients (Figure 1). The median tumor size (range) was 17.5 (12–22) mm. Well-defined, clear rims were observed in five patients. Since we did not rule out malignant testicular tumors, we performed high inguinal orchiectomy in all patients. All tumors were macroscopically well-defined, white-toned masses (Figure 2). Microscopically, they were cysts with thin fibrous capsules. The inner surface of each cyst was covered with stratified squamous epithelium, and the inside was filled with keratinized substances (Figure 3). There were no skin appendages or other tissue components inside the cyst. All patients were diagnosed with testicular epidermoid cysts.

Discussion

Testicular epidermoid cysts, first reported by Dockerty et al. in 1942, are benign tumors that account for 1% of all testicular tumors.¹ They commonly affect patients 10–39 years of age, which was almost similar to the susceptible age of having germ cell tumors (between 20 and 39 years). Epidermoid cysts are classified as prepubertal teratoma, but we have to realize that they frequently occur in adulthood.^{2,5} Price et al. reported that 87% of all testicular epidermoid cysts were small tumors of <30 mm in size.² According to the 2016 WHO classification, epidermoid cysts are classified as prepubertal teratomas unrelated to germ cell neoplasia in situ (GCNIS). Postpubertal teratomas are considered malignant because they are derived from GCNIS. Contrarily, prepubertal teratomas arise from normal primordial embryos which makes them, including epidermoid cysts, benign lesions.⁶

More than 70% of pediatric testicular tumors are benign; therefore, testicular-sparing surgery is often performed.⁷ The prerequisites for the tumor to be considered benign prior to testicular-sparing surgery are particular US findings (cystic lesions with clear borders and calcified contents) and AFP negativity. In addition, it is necessary to confirm that there are no malignant findings on intraoperative frozen section examination (FSE).^{8,9} We encountered one pediatric patient, but because the boundary was relatively ill-defined on US findings (Figure 1C), high inguinal orchiectomy was performed, considering the risk of malignant dissemination after tumor enucleation alone.

Testicular epidermoid cysts are recognized as benign tumors, and there have been no reports of their metastasis. A report of 69 cases of testicular epidermoid cysts worldwide showed no metastases or death from this disease during the 1-month to 24-year observation periods.¹ In the present report, no recurrence was observed during the observation period of 2 weeks to 3 years.

Testicular tumor markers were negative in patients with epidermoid cysts. Previous studies have also indicated that 80% of small tumors (less than 20–25 mm, regardless of age) are benign, and small-sized tumors are a predictor of benign tumors.^{10–12} In this study, all patients were negative for tumor markers, and their tumors were within 25 mm in size.

US is highly useful for making preoperative diagnoses. Typically, the outer edge has a hyperechoic partition, called an echogenic rim. A low-echo level concentric ring (represented as an onion ring or a bull's eye) is seen inside when the alternating layers of compressed keratin and exfoliated squamous epithelial cells are regular. However, when the layers are randomly arranged with a slightly alternating pattern, the lesion looks like a heterogeneous mass and can present varied findings. Reflecting the internal keratinization, it is avascular in Doppler US.^{13,14} In five of our six patients, hyperechoic septa were confirmed on the outer edges and well-defined tumors were visualized.

For histological diagnosis, Price et al. reported four diagnostic criteria: first, the cyst wall exists in the parenchyma of the testis; second, the inside contains layers of keratinized and non-structural substances; third, the cyst wall surrounds the stratified squamous epithelium; and fourth, an epidermoid cyst is defined

as having no teratoma-like tissues or skin appendages consisting of fibrous connective tissues.²

Recently, testicular-sparing surgery has begun to be recognized to avoid the overtreatment of benign lesions and preserve sexual functions. According to the European Association of Urology clinical guidelines (2022), high inguinal orchiectomy without intraoperative FSE should not be performed in patients with suspected benign testicular tumors.¹⁵ Fankhauser et al. reported the accuracy of intraoperative FSE for testicular tumors.¹⁶ Axel et al. performed testicular-sparing surgeries in 18 patients with testicular epidermoid cysts. All patients were accurately diagnosed as having epidermoid cysts by FSE.⁴

Conclusion

In this study, we reported six cases of rare testicular epidermoid cysts. When encountering patients with negative tumor markers, clear margins, negative Doppler flow, and sole small testicular tumors ([?]25 mm), we should always consider the possibility of benign epidermoid cysts to avoid overtreatment.

Author Contributions

Kimitsugu Usui: Conceptualization; data curation; writing – original draft. Ryo Yamashita: Conceptualization; data curation; project administration; writing – review and editing. Yuma Sakura: Conceptualization; data curation; project administration; writing – original draft. Masafumi Nakamura: Data curation; writing – review and editing. Hideo Shinsaka: Data curation; writing – review and editing. Masato Matsuzaki: Data curation; writing – review and editing. Masashi Niwakawa: Project administration; supervision; writing – review and editing.

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Conflict of interest statement

The authors declare no conflict of interest.

Data availability statement

The authors confirm that the data supporting the findings of this study are available within the article.

Ethics statement

The protocol for this research project has been approved by a suitably constituted Ethics Committee of the institution, and it conforms to the provisions of the Declaration of Helsinki. Institutional review board of Shizuoka Cancer Center, Approval No. 3625.

Consent

The study information was displayed on the hospital website homepage to provide a means for patients to opt out.

Registry and the Registration No. of the study/trial

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Shizuoka Cancer Center Clinical Research Ethics Review Committee

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Figure legends

Figure 1 Ultrasonography of the testicular tumor. Well-defined, clear rims are observed in all cases except C.

Figure 2 Macroscopic appearance of epidermoid cyst of the testis. A round white nodule with a keratinized material inside is seen.

Figure 3 Microscopic appearance of an epidermoid cyst of the testis. The cystic mass is filled with keratinized material. The wall is composed of dense fibrous tissues lined by stratified squamous keratinized epithelium.

Table 1 Characteristics of the six patients with testicular epidermal cysts

	Age (years)	Side	Chief complaint	Tumor size (mm)	Tumor marker levels	Figure
Case 1	27	Left	Scrotal induration	22	Normal	A
Case 2	19	Left	Scrotal induration	18	Normal	B
Case 3	9	Right	Scrotal induration	17	Normal	C
Case 4	21	Left	Scrotal induration	15	Normal	D
Case 5	30	Left	Pruritus scrotum	20	Normal	E

	Age (years)	Side	Chief complaint	Tumor size (mm)	Tumor marker levels	Figure
Case 6	18	Right	Scrotal induration	12	Normal	F

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