

CASE REPORT

Epidermoid cyst of the testis: A report of three cases

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Key Clinical Message

Testicular epidermoid cysts have characteristic findings. Testicular tumor markers are negative in patients with epidermoid cysts. Clear margins and sole and small testicular tumors (20 mm or less) suggest the possibility of epidermoid cyst. Testicular-sparing surgery with intraoperative frozen section examination should be performed when suspecting epidermoid cysts. Testicular epidermoid cysts are rare tumors that account for 1% of all testicular tumors and are often clinically misdiagnosed as malignant lesions. We report three cases of epidermoid cysts. The chief manifestations were scrotal induration in two patients and pruritus scrotum in one. The median age of the patients was 23 years (18–30). All tumors were determined to be sole lesions (<20 mm in diameter). Testing for tumor markers in all patients revealed negative results. We could not rule out malignancy; hence, we performed high inguinal orchiectomy in all cases. Histologically, the inner walls of the cysts were lined with stratified squamous epithelium; their contents were keratinized. All patients were diagnosed with epidermoid cysts.

KEYWORDS

epidermoid cyst, orchiectomy, testicular neoplasms, ultrasonography

1 | INTRODUCTION

Testicular epidermoid cysts are rare tumors that occur in approximately 1% of all patients with testicular tumors and are often found in young men aged 10–39 years.^{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 establish. 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. In recent years, testicular preservation therapy including rapid intraoperative diagnosis

of benign testicular tumors has become popular. We summarized specific preoperative findings differentiating epidermoid cysts from malignant lesions. Herein, we report three rare cases of epidermoid cysts in the testes.

1.1 | Case history/examination

We encountered three patients with epidermoid cysts at our hospital between April 2015 and March 2022 (Table 1). The median age (range) was 23 (18–30) years. The tumors occurred in the right testes in one patients and the left in the other two. The chief manifestations were the presence

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of a scrotal mass in two patients and scrotal itching in one. 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 15 (12–20) mm. Well-defined, clear rims were observed in all patients.

1.2 | Differential diagnosis, investigations, and treatment

Since we did not rule out malignant testicular tumors, we performed high inguinal orchiectomy in all patients.

1.3 | Outcome and follow-up

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; 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.

2 | DISCUSSION

Testicular epidermoid cysts, first reported by Dockerty and Priestley in 1942, are benign tumors that account for

1% of all testicular tumors.¹ They commonly affect patients at 10–39 years of age, which was almost similar to the susceptible age of having germ cell tumors (between 20 and 39 years). Although epidermoid cysts are classified as prepubertal teratoma, we have to realize that they frequently occur in adulthood.^{2,5} Price 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 that make them, including epidermoid cysts, benign lesions.⁶

Testicular epidermoid cysts are recognized as benign tumors; there have been no reports on 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

Testing for testicular tumor markers revealed negative results 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.^{7–9} In this study, all patients were negative for tumor markers, and their tumors were within 20 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. Manning and Woodward reported that concentric ring pattern was observed in 62% of cases. However, when the layers are randomly arranged with a slightly alternating pattern, the

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 | 21 | Left | Scrotal mass | 15 | Normal | A |
| Case 2 | 30 | Left | Scrotal itching | 20 | Normal | B |
| Case 3 | 18 | Right | Scrotal mass | 12 | Normal | C |



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

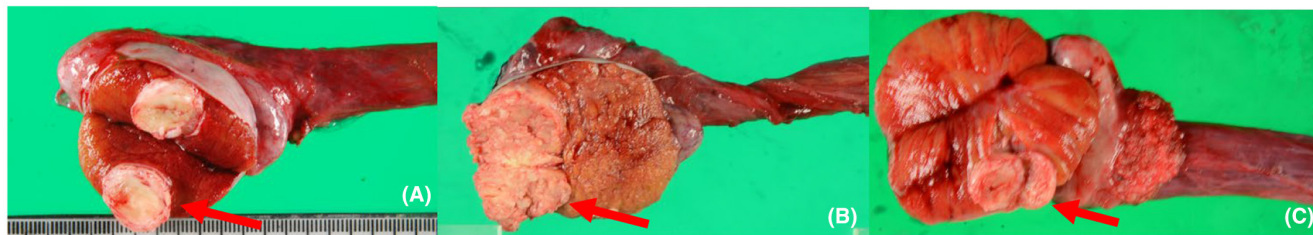


FIGURE 2 Macroscopic appearance of epidermoid cyst of the testis. A round white nodule with a keratinized material inside is seen.

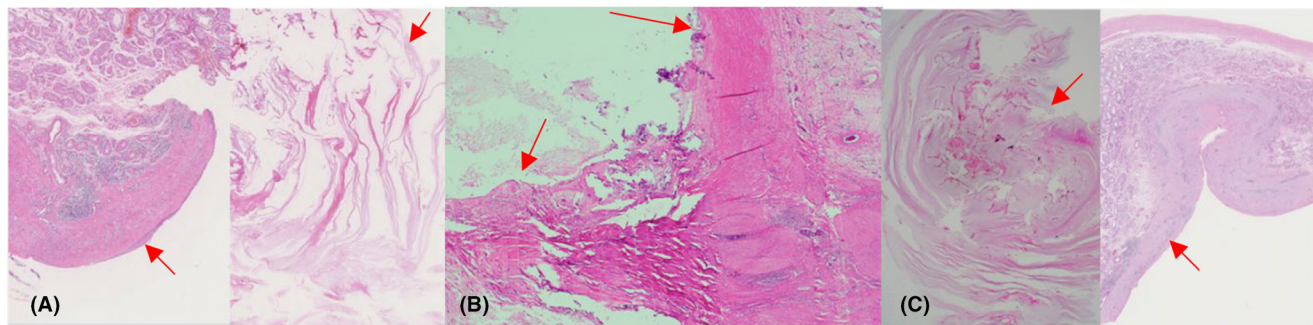


FIGURE 3 Microscopic appearance of an epidermoid cyst of the testis. The cystic mass is filled with keratinized material. The wall is composed of dense fibrous tissues lined by stratified squamous keratinized epithelium.

lesion looks like a heterogeneous mass and can exhibit varied findings. Reflecting the internal keratinization, it is avascular on Doppler US.^{10–12} In all patients, hyperechoic septa were confirmed on the outer edges and well-defined tumors were visualized.

Regarding histological diagnosis, Price 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.¹³ Fankhauser et al. reported the accuracy of intraoperative FSE for testicular tumors.¹⁴ Heidenreich et al. performed testicular-sparing surgeries in 18 patients with testicular epidermoid cysts. All patients were accurately diagnosed with epidermoid cysts by FSE.⁴ However, in recent reports, there have been cases in which the final diagnosis was germ cell tumor in the surrounding area in cases wherein the cyst was thought to be an epidermoid cyst based on preoperative images and intraoperative

diagnosis; orchidectomy was additionally performed. Establishing a 100% accurate preoperative diagnosis is difficult.¹⁵

3 | CONCLUSION

In this study, we report three 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. Furthermore, surgeons should offer the treatment of testicular-sparing surgery with intraoperative FSE to patients.

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.

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