CASE REPORT

Pilomatrixoma-like testicular tumour without teratomatous elements

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SUMMARY

A 40-year-old man presented to us with infertility. During scrotal ultrasonography, a heterogeneous nodular mass was seen in the right testicle. Surprisingly, histopathological examination revealed shadow cells, which are the characteristic feature of pilomatrixomas.

BACKGROUND

Pilomatrixomas are found primarily in the maxillo-facial region and on the arm, leg or trunk, in order of frequency.^{1 2} No case of testicular pilomatrixoma has been published. Three cases in the literature have reported shadow cells in the testis. The first was a pilomatrixoma-like dermoid cyst.³ The second was a mature cystic teratoma.⁴ The third case shared similar histological findings, but the presence or absence of teratomatous elements accompanying the mass was not noted.⁵ To the best of our knowledge, this report describes the first documented case of a testicular tumour similar to ordinary pilomatrixoma, without teratomatous elements.

CASE PRESENTATION

Scrotal ultrasonography of a 40-year-old man who reported of infertility showed a 16×12 mm heterogeneous nodular mass in the superomedial part of the right testicle, within the parenchyma. Atrophic appearance and grade 3 varicocele were detected at the left testicle. In scrotal MRI, a hypointense rim was observed with all sequences at the periphery of a nodular mass with a smooth border (figure 1). Levels of tumour markers such as α -fetoprotein, β -human chorionic gonadotropin and lactate dehydrogenase were within normal limits. Although the patient reported infertility, motile sperm was found in a semen analysis. Since the patient presented with a small-diameter tumour and atrophy in the other testicle, intraoperative evaluation of frozen sections was planned. The multidisciplinary treatment team decided to perform partial orchiectomy, if no findings of malignancy were noted intraoperatively. After receiving informed consent from the patient, surgery was performed. In histopathological evaluation of frozen sections, no malignant findings were noted, and partial orchiectomy was performed. Histopathological examination of sections of the complete specimen revealed a cystic lesion with clear borders, without a capsule. Within the lesion, in addition to calcification embedded in fibrotic stroma and extensive ossification, shadow cell groups were also seen (figures 2 and 3). No

Figure 1 Coronal T2-SPIR (selective partial inversion recovery) MR imaging; more hyperintense lesion than testicular parenchyma, having hypointense ring with well-defined borders at the superior of the right testicle is seen.

teratomatous elements or viable squamous epithelial cells including the cutaneous adnexa were observed, and focal spermatogenesis was identified in testicular tissue adjacent to the lesion. A diagnosis of pilomatrixoma-like tumour was performed. After surgery, a chest X-ray and abdominal/pelvic CT scan were performed, and no extratesticular involvement was detected. In 2 years of follow-up, there was no recurrence.

DISCUSSION

Pilomatrixoma is a benign tumour of cutaneous appendages with differentiation towards the matrix of the hair follicle. The reported incidence of pilomatrixoma is 0.08% in surgical pathology cases and 1.04% when benign dermatological lesions are included.¹

Figure 2 Testicular tumour contains calcified and ossified areas (H&E ×40).

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Figure 3 In addition to ossification, large amounts of eosinophilic and anuclear shadow cell groups which are characteristic findings of pilomatrixoma (H&E $\times 200$ (A), H&E $\times 100$ (B)).

The aetiology is not known; a relationship to trauma, insect bite or surgery was noted in the history of fewer than 5% of cases.¹

Histopathologically, pilomatrixoma consists of two main cell types, basophilic cells and eosinophilic shadow cells. The basophilic cells tend to be at the periphery of the cell island, and have little cytoplasm, indistinct cell borders, hyperchromatic nuclei and plentiful mitoses. The eosinophilic shadow cells are usually found nearer the centre of the cell mass. They have more cytoplasm and distinct cell borders, but no nuclear staining. It has been suggested by some authors that shadow cells form from the apoptosis of basophilic cells.⁶

According to histopathological findings, pilomatrixomas are chronologically divided into four phases: early, fully developed, early regressive and late regressive. In the late regressive period, shadow cells, calcification, ossification and fibrotic stroma develop, and a complete loss of the basaloid epithelial component is observed.¹ Our case, by these criteria, shows similarity to late regressive period pilomatrixomas.

The differential diagnosis is primarily with dermoid cyst. Testicular dermoid cyst is a specialised form of mature teratoma, in which cysts lined with squamous epithelium and accompanying cutaneous adnexa are observed. Between these cutaneous adnexa, hair follicles and sebaceous glands are present. Dermoid cysts may contain other teratomatous elements in small areas.⁷ Fewer than 20 cases of testicular dermoid cyst have been reported. In our case, in addition to calcification in fibrotic stroma and extensive areas of ossification, shadow cell groups were seen. Teratomatous elements or viable squamous epithelial cells including the cutaneous adnexa were not seen. This histopathological appearance is different from that of a dermoid cyst. Pilomatrix carcinoma must also be considered in the differential diagnosis. Pilomatrix carcinoma is composed of pleomorphic basaloid cells with prominent nucleoli and frequent mitoses. Necrosis is sometimes present. Keratotic material and shadow cells may be present in the centre of basaloid islands. We saw neither basaloid cells nor necrosis in histopathological examination of sections of the complete specimen from our case. Also, it should be remembered that shadow cell differentiation can occur in some visceral malignancies, such as endometrial and colon adenocarcinomas with squamous cell differentiation.⁶ Both of these tumours appear morphologically as adenocarcinomas, and their epithelial components show characteristic features of structural and cellular atypia, as well as frequent mitoses, necrosis and invasion. We saw none of these in the present case.

In the literature, there have been three similar cases localised to the testicle. In one case, eosinophilic shadow cells, peripheral calcification and ossification were observed in the central part of a dermoid cyst, which was identified as a pilomatrixoma-like dermoid cyst.³ In another case, in addition to shadow cells, calcification and ossification were observed. In this report, the presence or absence of other teratomatous elements accompanying the mass was not mentioned.⁵ In the third case, nest-forming squamous epithelium was seen between the shadow cells and calcified, ossified structures were observed. Also, within a fibrous pseudocapsule, smooth muscle bundles were present.⁴ This was a mature cystic teratoma. In our case, there were no teratomatous elements such as squamous epithelium or smooth muscle.

In adults, the treatment for all testicular germ cell tumours is radical inguinal orchiectomy. The organ-sparing, frozen section analysis-guided approach to small testicular masses (25 mm or less in diameter) represents a treatment alternative to orchiectomy in selected patients with tumours in a solitary testis. Organ-sparing surgery aims at preserving enough testicular parenchyma to maintain physiological endocrine function and, if possible, fertility.⁸ Since in our case the patient's report was of infertility, and as no malignant development was identified during surgery, a partial orchiectomy was performed. The patient is undergoing follow-up at the time of writing, and no problems have been observed.

Learning points

- ▶ Histopathologically, pilomatrixoma consists of two basic cell types, basophilic cells and eosinophilic shadow cells.
- ▶ In the late regressive period, the basophilic cells can be completely lost and one may see only eosinophilic shadow cells.
- ▶ Differential diagnosis is mainly with dermoid cyst. Testicular dermoid cyst is a specialised form of mature teratoma, in which cysts lined with squamous epithelium and accompanying cutaneous adnexa are observed.
- ▶ To the best of our knowledge, this report describes the first documented case of testicular tumour similar to ordinary pilomatrixoma, without teratomatous elements.

Competing interests None.

Patient consent Obtained.

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