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CASE REPORT

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Angiosarcoma of the testis

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Abstract A primary angiosarcoma of the testis in a 74-year-old patient was a highly anaplastic epthelioid angiosarcoma, which was positive for endothelial markers immunohistochemically. The tumour was unrelated to testicular germ cell neoplasm; the patient had received no previous radiation or chemotherapy.

Key words Testis · Angiosarcoma · Germ cell tumours

Introduction

Angiosarcoma of the testis is an extremely rare tumour; the one case reported developed within a well-differentiated testicular teratoma [2]. Two other documented angiosarcomas associated with testicular germ cell tumours arose in retroperitoneal metastases and in the gut, following radiation and chemotherapy [4, 5].

Case report

A 74-year-old male was admitted with an intermittent fever of 3 weeks duration. Routine clinical check-up failed to reveal its cause. Ultrasonography showed a transonic, well-vascularised lesion in the left testis, measuring 11×17×11 mm. After orchiectomy the patient became afebrile and the previously elevated erythrocyte sedimentation rate (ESR) and C-reactive protein returned to normal values. Pathological examination revealed a tumour in the left testis, which was histologically an epithelioid angiosarcoma. No regional or distant metastases were detected by thorough clinical examination. One month later the patient died at home of a stroke. No autopsy was performed.

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Materials and methods

For light microscopy the tissue specimens were routinely processed and stained with H&E, and with silver impregnation for reticulin. Immunohistochemically, the paraffin-embedded 4-µm-thick sections were processed with the avidin–biotin complex (ABC) immunoperoxidase method using the antibodies to vimentin, factor VIII-associated antigen, CD 31, LCA, PLAP, CD 68, cytokeratin (all DAKO), CD 34 (Becton-Dickinson), and Ulex europaeus lectin (EY Lab) with DAB as a chromogen. For electron microscopy the specimens were prepared from paraffin sections, fixed in OsO_4 and embedded in Epon 812. Ultrathin sections were stained with uranyl acetate and lead citrate. Image cytometric DNA content analysis of the tumour cell nuclei was also performed.

Pathology

The submitted left testis and attached 6-cm portion of spermatic cord weighed 50 g. In the central part of the testis a poorly delineated tumour mass was found, measuring 13 mm in its largest diameter. The cut surface of the tumour was slightly mottled and brownish white in appearance.

Histologically, the tumour had infiltrated widely into the adjacent testicular tissue and entrapped numerous seminiferous tubules (Fig. 1). The tumour cells had the appearance of highly atypical endothelial cells, arranged somewhere along the lumina of poorly formed anastomosing vascular spaces (Fig. 2). The tumour cells were polygonal, rounded or elongated, with indistinct boundaries and strongly eosinophilic cytoplasm. Large vesicular nuclei with irregularly dispersed chromatin were found, showing conspicuous nucleoli and vivid mitotic activity. Focally, a moderate lymphocytic infiltrate was observed in the stroma, as well as discrete collections of haemosiderin.

Immunohistochemically, positive reactions for vimentin, factor VIII-related antigen, CD 31, CD 34 and Ulex europaeus lectin suggested an endothelial cell origin of the tumour (Fig. 3). Immunohistochemical reactions on cytokeratin, CD 68, LCA and PLAP were negative. Elec-

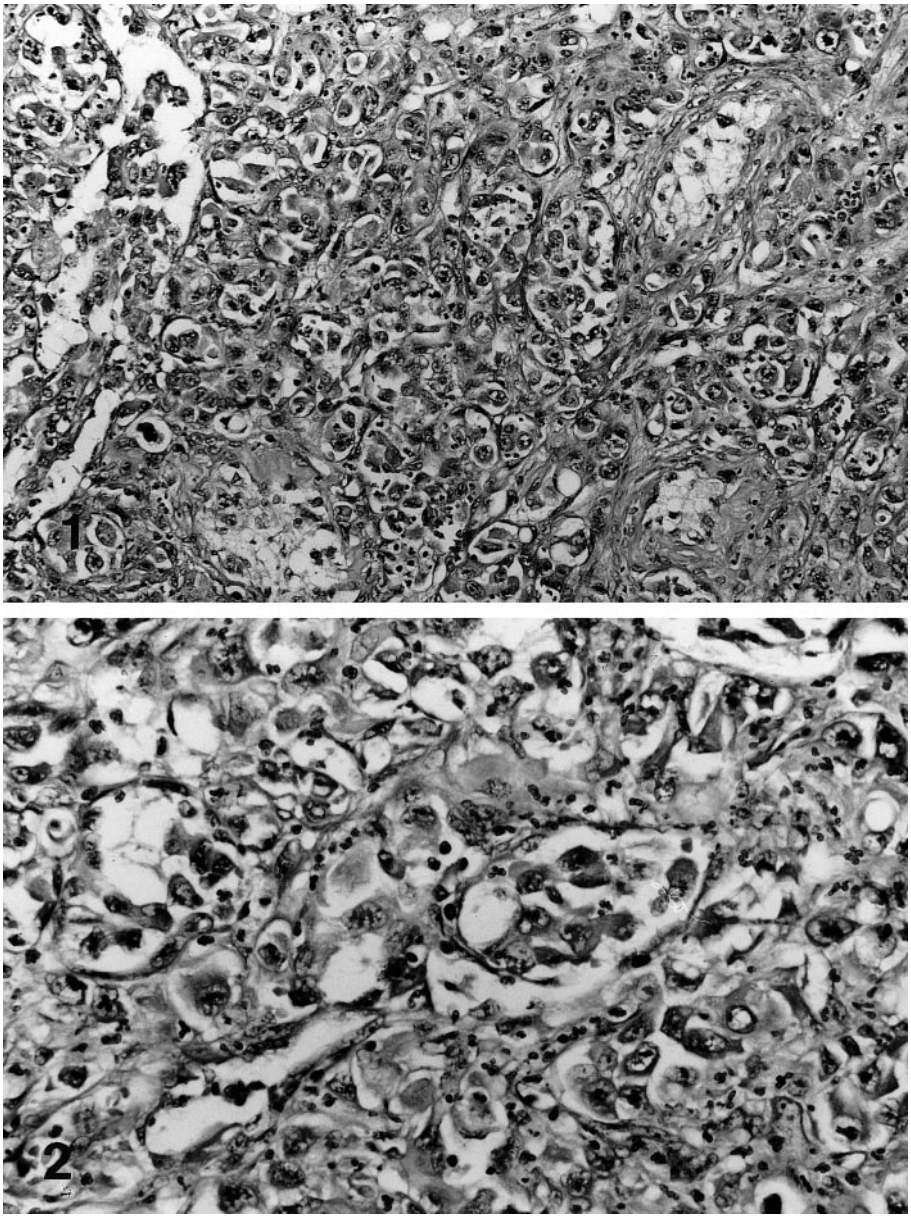

Fig. 1 Epithelioid angiosarcoma with some residual seminiferous tubules. H&E, ×175

Fig. 2 Pleomorphic tumour cells with prominent nucleoli. H&E, ×350

tron microscopy examination revealed characteristic Weibel-Palade bodies and occasional erythrocytes in the cytoplasm of some tumour cells (Fig. 4a, b). The image cytometric DNA content of the tumour cell nuclei showed an aneuploid peak (DI=1.82).

Discussion

Angiosarcoma is an uncommon neoplasm that can develop at various sites including skin, soft tissues, breast, liver, bone, and spleen [1]. However, its occurrence in the testis seems to be exceedingly rare [2, 5]. Hughes et al. [2] described a case of testicular angiosarcoma arising within a well-differentiated teratoma in a 16-year-old patient. Angiosarcoma was also reported in the retroperitoneal metastases of a mixed germ cell tumour in a patient who had been treated with radiation and chemotherapy [5]. This mode of therapy may be implicated in the development of angiosarcoma associated with germ cell tumours [3–5]. It is well known that non-germ-cell tumours can also develop in teratomas without previous therapy. Well-documented cases of carcinoids, nephroblastomas and primitive neuroectodermal tumours in the testis have been reported [6]. In all these cases, small teratomatous components and/or atypical germ cells in the preserved tubuli were found. These phenomena were explained by the fact that a single somatic component of the teratoma had overgrown all other constituents. According to Young and Scully [6], such tumours are classified as “monodermal and highly specialised” teratomas.

In our case, the entire surgical specimen was examined histologically but neither a teratomatous component

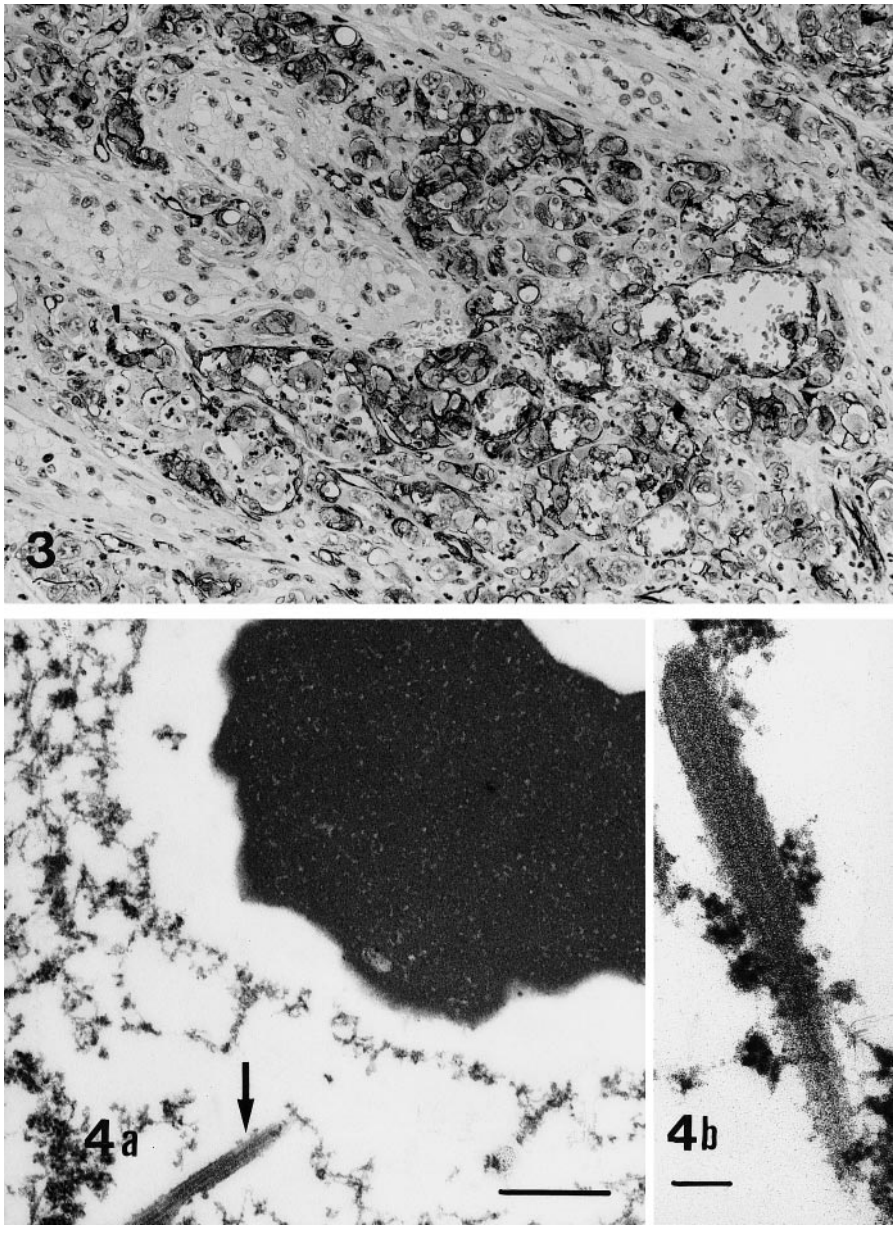

Fig. 3 Tumour cells showing strong immunoreactivity for CD 31, ×175

Fig. 4 **a** An erythrocyte and Weibel-Palade body (*arrow*) in the cytoplasm of the tumour cell. *Bar* 0.5 µm **b** Weibel-Palade body at higher magnification. *Bar* 0.1 µm

nor atypical germ cells (intratubular germ cell neoplasia) were found. Moreover, the patient had not any previous chemo- or radiotherapy. Therefore, we can assume that the present tumour is a primary angiosarcoma not related to the germ cell neoplasia.

The prognosis of testicular angiosarcoma is probably poor, although in our case we are not able to contribute any information in this regard; the patient died of an unrelated cause.

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