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Oncology

Rare case of squamous cell carcinoma arising from tunica vaginalis in a background of chronic hydrocele

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Introduction

Paratesticular region tumors are uncommon and include epithelial, mesothelial, and mesenchymal derived lesions. While adenomatoid tumor is the most common benign paratesticular tumor, malignant mesothelioma is a very rare but well documented occurrence.[1] Liposarcoma and rhabdomyosarcoma are the most common malignant paratesticular tumors in adults and children, respectively. Other primary epithelial tumors that can arise from this area include benign and malignant rete testis lesions and ovarian-type epithelial tumors.[1] We report a case of SCC arising from the tunica vaginalis in the context of chronic hydrocele. To the best of our knowledge, SCC in paratesticular region has been reported twice in the English literature.[2,3]

Case presentation

A 76 year old male with long-standing history of hydrocele presented for evaluation of right testicular pain and swelling. Imaging studies of the scrotum revealed a large, malignant appearing mass arising from the right testis. An inguinal orchiectomy revealed a 4.5 cm tan tumor in the lower pole of the testis.

Histopathological examination revealed a tumor composed of large nests and thick cords of squamous epithelioid cells with eosinophilic, amphophilic and clear cytoplasm with well demarcated cell borders, intercellular bridges, and focal keratinization (Fig. 1 A, B, C). The cells contained large pleomorphic nuclei with vesicular chromatin and prominent nucleoli. Lymphovascular invasion was identified with associated necrosis and frequent mitotic figures. The tumor abuts the adjacent normal testicular parenchyma with no evidence of infiltration (Fig. 1D, arrows). Dartos muscle was involved and there was no connection of the tumor to overlying epidermis. Histomorphologic features that would suggest either a teratoma or a malignant mesothelioma were not encountered following extensive sampling of the lesion. No evidence of Germ Cell Neoplasia in Situ (GCNIS) was noted in the adjacent testis.

A cystic structure with calcifications and fibrosis consistent with long-standing hydrocele was identified (Fig. 1D, arrowheads). The hydrocele lining is predominantly composed of a mesothelial monolayer with focal areas of atypical cuboidal cells with increased mitotic activity (Fig. 1E). The tumor appeared to be arising from the cystic lining (Fig. 1F, arrows).

Ber-Ep4 showed patchy immunoreactivity in the tumor (Fig. 2A) and hydrocele lining (Fig. 2B). The tumor was diffusively immunoreactive for Pancytokeratin, CK7 and p63 (Fig. 2C). Both Calretinin (Fig. 2D) and PLAP were also immunoreactive, with the former showing focal positivity and the latter showing strong and patchy positivity, particularly in the metastatic deposit. CK20, p16, PSA, OCT3/4, WT1, SOX10, TTF1, adipophilin, androgen receptor, and CDX2 were not reactive. Interestingly, the hydrocele lining showed focal expression for Ber-Ep4, p63 and Calretinin while being negative for WT1, further suggesting a potential origin for the SCC.

The tumor was diagnosed as invasive poorly differentiated SCC involving paratesticular tissue with associated hydrocele. Extensive work up and review of the patient medical history failed to identify an extra

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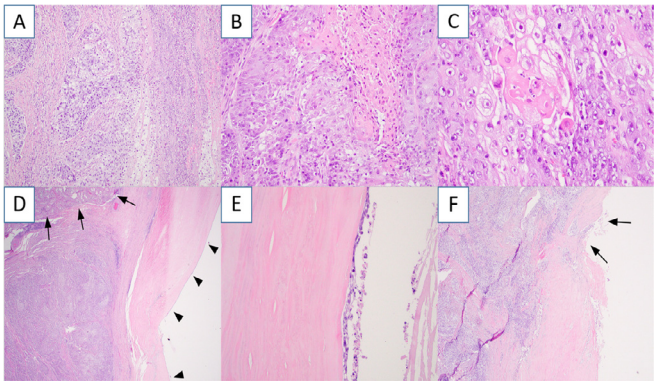

Fig. 1. Composite image displaying the histomorphological features of the main tumor. **(A)** Low power view of the tumor showing thick cords and nests of cells with eosinophilic to clear cytoplasm and necrosis. **(B & C)** Mid and high power view highlighting foci of keratinization **(D)** Low power magnification showing tumor abutting benign testis (arrows) and immediate proximity to the cyst (arrowheads) **(E)** High power magnification of the cyst lining, showing atypical flat to cuboidal cells **(F)** Mid and high power magnification showing the tumor seemingly emerging from the cystic wall (arrows) (Hematoxylin and Eosin).

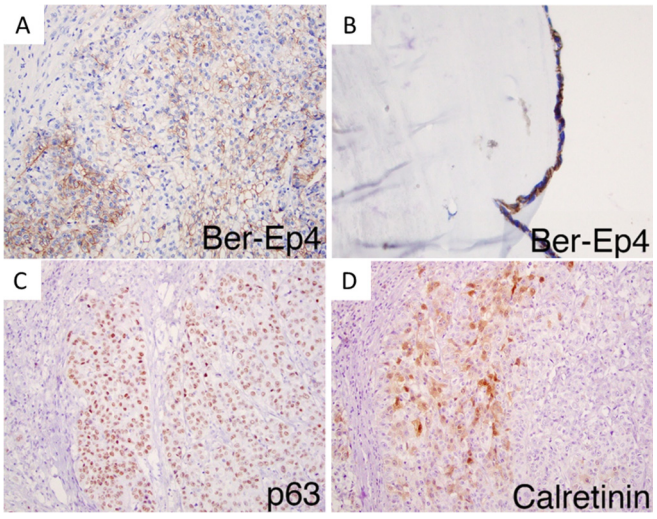

Fig. 2. Composite of immunohistochemical features of the main tumor and cyst lining. **(A & B)** Membranous positivity for Ber-Ep4 on main tumor and cystic lining cells. **(C)** Nuclear positivity for p63 and **(D)** Focal immunoreactivity for Calretinin in main tumor.

testicular primary tumor. This with the above described findings strongly support a primary paratesticular SCC.

Adjuvant radiation and chemotherapy were administered given the involvement of tumor resection margins. The patient developed metastatic disease in ipsilateral inguinal lymph nodes 2 months following initial diagnosis. The metastatic tumor revealed identical SCC morphology with keratinization (Fig. 3A and B).

Discussion

We describe an extremely rare occurrence of an aggressive paratesticular SCC most likely originating from the mesothelial lining of the tunica vaginalis in association with a long-standing, chronic hydrocele. The histologic and immunoprofile of the tumor is most consistent with SCC as supported by the strong and diffusive positivity for CK7, p63, Pancytokeratin and Ber-EP4 and the presence of frank keratinization in metastatic deposits.

In our case, given the association of a long-standing hydrocele, a primary mesothelioma was considered and ruled out. Classic morphologic features of mesothelioma were absent and markers of mesothelial differentiation (e.g. Calretinin) were positive only in the hydrocele lining but primarily negative in the associated tumor.

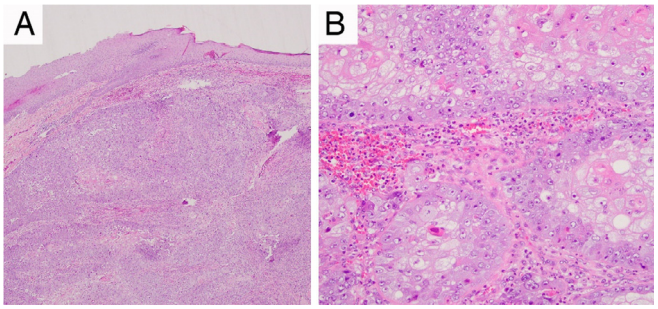

Fig. 3. (A & B) Low to high power Hematoxylin and Eosin stains on the inguinal metastatic deposits, showing same histomorphologic features as the main tumor.

In this location, the differential diagnosis of a SCC includes: i) teratoma with somatic-type malignancy; ii) metastasis of SCC from extra testicular sites; iii) secondary extension of a scrotal or skin adnexal primary SCC, iv) and least likely a paratesticular region primary SCC. Teratomas can seldom demonstrate a somatic-type malignant component and could account for paratesticular SCC. Our case did not have morphologic or immunohistochemical (OCT3/4 negative) features of teratoma or other germ cell tumors and lacked the presence of GCNIS. Metastases to paratesticular region are rare. As indicated above, clinical exploration failed to show any other primary tumor. Extension from a primary scrotal skin or skin adnexal origin was also ruled out in our case given the lack of involvement of the overlying skin and negativity for immunomarkers of adnexal differentiation (e.g. adipophilin and androgen receptor).

To our knowledge, two other cases of primary paratesticular SCC have been reported. Bryan et al. describe a SCC arising in association with chronic hydrocele,[2] similar to our case. Artemyeva et al. describe a “low-grade non-keratinized squamous cell carcinoma of the left testicle with metastasis to the retroperitoneal lymph nodes and the lungs” with no evidence of teratoma or presence of tumor in other sites outside the scrotum.[3]

In our tumor, the patchy immunoreactivity for calretinin and PLAP (a marker that can be positive in a subset of mesotheliomas[4]), but not other mesothelial markers such as WT1 and gain of squamous epithelial markers could be interpreted as supportive of oncogenic transformation from mesothelial lining of the hydrocele. Furthermore, the above described focal gain of expression for Ber-Ep4 and p63 in the hydrocele is also supportive of a metaplastic process in a setting of long standing chronic inflammation. Interestingly, SCC transformation (or divergent differentiation) of mesothelioma was recently documented in a thoracic mesothelioma in which there was complete loss of mesothelial markers in the SCC component.[5]

Conclusion

We report the third case of primary SCC of the paratesticular region. Although a rare occurrence, this possibility should be taken into account in cases of rapidly growing tumors in the context of chronic hydrocele.

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Declarations of interest

None.

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