CASE REPORT

Sarcomatoid Squamous Cell Carcinoma of the Penis – a Report of Two Cases

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Background: Sarcomatoid (spindle cell) squamous cell carcinoma is a rare, high-grade, aggressive tumor consisting of the squamous cell carcinoma admixed with the malignant spindle cell (sarcomatoid) elements. These tumors are relatively uncommon in the genitourinary system and particularly in the penis.

Materials and methods: Two sarcomatoid squamous cell carcinomas of the penis were diagnosed in our hospital between 2012 and 2015. Clinical histories, pathology reports, hematoxylin and eosin-stained and immunohistochemical slides were reviewed.

Results: In both cases, the tumors presented as single, pedunculated, extensive masses with surface ulceration; histology study showed a mixture of high-grade squamous cell carcinoma component and spindle cell neoplastic component in different proportions. Immunohistochemical stains of CK AE1/AE3, p63 and CK903 showed positive immunoreactivity in both components in both cases. Vimentin was positive in spindle cell component and negative in squamous cell carcinoma areas.

Conclusion: Sarcomatoid squamous cell carcinoma of the penis is an uncommon tumor of this site with aggressive behavior and bad prognosis which might be related to the delay in medical examination and diagnosis. A correct and thorough morphological study is of great importance for the staging of the disease, treatment and follow up of patients.

BACKGROUND

Sarcomatoid (spindle cell) squamous cell carcinoma is a rare, high-grade, aggressive tumor consisting of the squamous cell carcinoma admixed with the malignant spindle cell (sarcomatoid) elements.¹ These tumors are relatively uncommon in the genitourinary system and particularly in the penis.² There are only two small studies and a few case-reports presenting this rare entity in which the authors discuss the potential origin of the tumor, the histological appearance and presumptive biological behavior.³⁻⁷ The current study adds two more cases complete with clinical and morphologic data that we collected.

MATERIALS AND METHODS

Between 2012 and 2015, we diagnosed two sarcomatoid squamous cell carcinomas of the penis

treated in our hospital. Clinical histories were taken from the patients' charts, and pathology reports or consultation with primary physicians. Hematoxylin and eosin-stained slides as well as immunohistochemical stains were reviewed.

RESULTS

CLINICAL MANIFESTATIONS

The age of the patients was 58 and 73 years. Both patients had a continuous disease history prior to the present diagnosis. In case 1, initially the patient noticed redness and peeling of the foreskin of the preputium and glans penis. Two years later, in the same area, a wart-like formation emerged, rapidly getting larger in the last two or three months. Clinical examination revealed an ulcerative mass with spontaneous bleeding and foul smell (**Fig. 1A**). In

case 2, the patient consulted a urologist because of bilateral hydrocele. Clinical examination established severe phimosis and a palpable node in the ventral part of the glans penis apart from the bilateral hydrocele (**Fig. 2A**). Both patients had no sign of palpable lymphadenopathy at the time of diagnosis; they underwent a surgical removal of the tumors. A summary of all clinical features of the patients is given in **Table 1**.

GROSS APPEARANCE

The tumors presented as a single, pedunculated,

extensive masses with surface ulceration. Sectioning revealed tan-white to gray color with firm texture masses. In case 2 the tumor infiltrated preputial skin which caused phimosis.

HISTOLOGIC FEATURES

Both tumors showed a mixture of high-grade squamous cell carcinoma component and spindle cell neoplastic component in different proportions. Case 1 had extensive squamous cell component with single areas of keratinization. Sarcomatoid elements of the tumor mostly resembled leiomyosarcoma composed

Table 1. Clinical characteristics of patients

Case	Age	History	Presentation	Diameter	Palpable lymph nodes	Distant metastasis	Surgery
1	58	3 years	Ulcerative mass	3,5 cm	No	No	Partial amputation
2	73	Few months	Hydrocele	2 cm	No	No	Partial amputation, excision of the hydrocele

Figure 1. (A) Penis showing extensive tumor formation with ulcerative surface (arrow) which occluded the entire glans; (B) Microscope picture of the tumor demonstrating two components – squamous (left side) and sarcomatoid (right side) (HE, x10); (C) Sarcomatoid component of the tumor showing moderate cytoplasmic expression of CK AE1/AE3 (x40); (D) Sarcomatoid component of the tumor showing strong cytoplasmic expression of vimentin (x40).

of spindle cells, which were closely situated and separated by collagen fibers with focal hyalinization (**Fig. 1B**). In case 2, most of the sarcomatoid areas showed spindle cells which “floated” in loose myxoid stroma (**Fig. 2B**). Both cases showed a typical tumor histology: the squamous cell carcinoma was superficially situated whereas the sarcomatoid areas had a prominent infiltrated pattern and affected penile tissue in deeper zones of the corpora spongiosa and cavernosa. **Table 2** shows a summary of the morphological features.

IMMUNOHISTOCHEMICAL ANALYSIS

CK AE1/AE3, p63 and CK903 showed positive immunoreactivity in both components in both cases (**Figs 1C, 2C**). Vimentin was positive in the spindle cell component and negative in the squamous cell carcinoma areas (**Figs 1D, 2D**). Additional CD 68, CD 45 and S-100 stains were diffusely negative in both tumor components. The human papillomavirus DNA status of the patients was not assessed since HPV DNA was found in neither of the cases previously.^{3,4}

Table 2. Morphologic characteristics of patients

Case	Tumor thickness	Depth of invasion	Surgical margins	Mitoses	Necrosis	Vascular invasion	Intrapanile metastasis
1	30 mm	Corpus spongiosum, tunica albuginea, corpus cavernosum	Negative	>5 HPF	Yes	Yes	No
2	13 mm	Corpus spongiosum, tunica albuginea	Negative	>5 HPF	No	Yes	No

HPF - high power field

Figure 2. (A) Penis showing nodular tumor formation (white arrow) which appears after resection of preputial skin of the glans (black arrow); (B) Microscope picture of the tumor demonstrating two components – squamous (right bottom) and sarcomatoid (left top) (HE, x10); (C) Squamous component of the tumor showing strong cytoplasmic expression of CK AE1/AE3 (x10); (D) Sarcomatoid component of the tumor showing strong cytoplasmic expression of vimentin (x40).

PROGNOSIS

A follow-up was conducted for both patients. The patient in case 1 underwent prophylactic bilateral inguinal lymph node resection three months after he was diagnosed, which was negative for metastasis. In case 2, sentinel lymph node resection was not performed because of advanced age and polymorbidity of the patients.

DISCUSSION

Sarcomatoid (spindle cell) squamous cell carcinoma is one of the morphological variants of the squamous cell carcinoma of the penis.² Histologically, sarcomatoid carcinomas appear as squamous cell carcinoma admixed in different proportions with spindle cell component resembling different types of sarcoma. This morphological heterogeneity was the reason why clinicians used a wide variety of terms to refer to this condition such as carcinosarcomas, biphasic tumor, and sarcomatoid carcinoma.⁶ Therefore, the question about the tumor's origin and nature arose in order to its better classification as well as patients' treatment and follow-up. Recent molecular and genetic studies establish monoclonal theory of the sarcomatoid carcinoma nature suggesting epithelial origin of both of the elements of the lesion despite some cases reporting cytokeratins' negative immunoreactivity particularly in the sarcomatoid component of the neoplasm.⁹ Other factors supporting this theory are mostly lymph node metastasis pathway of the tumor and the histological picture of squamous cell carcinoma detected in positive lymph nodes.^{3,4,8} Consequently, as of 2004 WHO recognized the term sarcomatoid carcinoma as the most appropriate one.¹⁰

Sarcomatoid carcinoma patients' age range widely (28 to 83 years) and the glans penis is very frequently the affected area.^{3,6} In most cases, patients had a progressive tumor development which correlates with our data³ and only one study reports an extremely rapid disease evolution⁴. We found a relation between the rate of development of the tumor and the underlying morphological characteristics. The rapidly growing lesions mostly exhibit predominance of the sarcomatoid component with minor to absolute absence of the squamous cell carcinoma elements. Conversely, a picture of the well-presented squamous cell carcinoma areas could be seen more often in slow progress tumors as in our cases.

Presently, the histological grade and level of the tumor invasion are considered as the most

important factors for prognosis of the disease local recurrence. The high-grade types of penile carcinoma with deeper invasion are associated with a high risk of the local recurrence.^{11,12} However, the ratio of the different components of the tumor had not been taken into consideration. Collection of a larger number of cases of sarcomatoid squamous cell carcinoma presenting different amounts of the squamous and spindle cell components with corresponding clinical, morphological and follow-up data might help for a better understanding of the biologic behavior of this tumor.

Morphologically, sarcomatoid squamous cell carcinomas should be distinguished from true sarcomas. There are some distinctive clinical, histological and molecular differences between them which could help in the differential diagnosis. While squamous cell carcinoma affects distal part of the penis and has an exophytic growth usually with superficial ulceration, true sarcomas arise from a deeper penile scarf.^{2,13} Microscopically, presence of the squamous cell carcinoma foci could be extremely helpful to make a diagnosis of sarcomatoid carcinoma. An absence of this component might require a total sampling of the specimen as well as multiple sections of the tissue especially close to the surface where squamous carcinoma areas could be found more frequently.³ Finally, immunohistochemical studies play an important role in detecting these carcinomas. Sarcomatoid carcinomas express diffusely or focally some keratins as CK AE1/AE3, CK903 or p63, EMA in both components of the tumor. Neoplastic spindle cells are usually strongly positive for vimentin and negative for SMA, desmin and S-100 protein.^{2,3,13}

CONCLUSION

Sarcomatoid squamous cell carcinoma of the penis is an uncommon tumor of this site with aggressive behavior and poor prognosis which might be due to the delay of the medical examination and diagnosis. An accurate, thorough morphological study is of great importance to the staging of the disease, the treatment and follow up of patients.

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Саркоматоидная сквамозная карцинома полового члена – сообщение о двух случаях

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Ключевые слова: саркоматоид-
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Введение: Саркоматоидная (веретенообразная) сквамозная карцинома является редкой, агрессивной опухолью высокой степени, состоящей из сквамозной карциномы, перемешанной со злокачественными веретенообразными (саркоматоидными) элементами. Данные опухоли встречаются сравнительно редко в мочеполовой системе и особенно в половом члене.

Материал и методы: В нашей больнице в период между 2012 г. и 2015 г. диагностированы два случая саркоматоидной сквамозной карциномы полового члена. Были рассмотрены клинические эпикризы, патологоанатомические доклады, предметные стекла, окрашенные гематоксилином и эозином и иммуногистохимические методы.

Результаты: В обоих случаях опухоль представляет собой одиночное обширное грибовидное образование с поверхностной ulcerацией; гистологические исследования демонстрируют смесь высокостепенного сквамозного карциномного компонента и веретенообразного неопластического компонента в различных пропорциях. Иммуногистохимические окрашивания СК AE1/AE3, p63 и CK903 раскрывают положительную иммунореактивность и в двух компонентах в обоих случаях. Виментин является положительным в веретенообразном компоненте и отрицательным в областях сквамозной карциномы.

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Заключение: Саркоматоидная сквамозная карцинома полового члена является редким случаем опухоли в данном местоположении с агрессивным поведением и плохим прогнозом, что может быть следствием запоздалого медицинского осмотра и диагноза. Компетентное и подробное морфологическое обследование является исключительно важным для определения стадии заболевания, лечения и контролирования пациентов.