Epithelioid Hemangioendothelioma Of The Penis: A Case Report

N Ghosh, T Jindal, M Kamal, R Sharma, P Gokulkrishnan, S Mukherjee, S Mondal, D Karmakar

INTRODUCTION

Epithelioid hemangioendothelioma is a rare vascular tumor. It is characterized by vessels lined by epithelioid endothelial cells along with inflammatory cell infiltrate. It can occur in different locations such as soft tissue, lung, liver, and bone. Penis is a very rare site for it. We report a case of penile epithelioid hemangioendothelioma and review the literature.

CASE REPORT

A 38 years old male presented with a non-healing ulcer over the glans penis for two months. His past history was unremarkable apart from a history of ceremonious circumcision at one year of age. On examination, there was an ulcerated lesion of size about 1 cm with irregular margins over the left side of glans penis close to corona (Figure 1).

The base was indurated but there was no tenderness. There was no inguinal lymphadenopathy. Clinically, carcinoma of penis was suspected hence an incisional biopsy was done from the margin of the lesion. Histopathological examination revealed focal ulceration of squamous epithelium with granulation tissue formation (Figure 2).
Epithelioid hemangioendothelioma is a rare vascular tumor that can occur in different locations such as soft tissue, lung, liver, and bone. Weiss and Enzinger initially described it in 1982 (1). It is a part of vascular proliferations characterized by the presence of epithelioid endothelial cells. It is considered a tumor of intermediate malignant potential between the benign variant called hemangioma and the more aggressive variant called epithelioid angiosarcoma. Epithelioid hemangioendothelioma most commonly involves the soft tissues and occurs as a solitary lesion. It can present with metastases. Rates of metastasis differ according to anatomical locations (20% in soft tissues, 25% in liver and 15% in lungs). Mortality rates also differ according to anatomical locations (13% in soft tissues, 35% in liver and 65% in lungs).

The penis is an unusual location for the disease. There are only eight papers in the English literature that describe the penile involvement. Most penile epithelioid hemangioendothelioma are solitary masses, although multifocal penile epithelioid hemangioendothelioma has been previously described in one report. It usually has an
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indolent course. Metastasis is rare.

There is no consensus regarding appropriate imaging for penile epithelioid hemangioendothelioma due to rarity of the cases. One study had used high frequency penile ultrasound to evaluate the lesion (3). Perhaps the first time, we have used Gadolinium contrast MRI to image the lesion – contrast enhanced MRI provided excellent anatomical details of the lesion as well as invasion of corporal bodies; which might be very important in decision making during surgical treatment.

Treatment of localized penile epithelioid hemangioendothelioma without distant metastasis includes wide local excision. Recurrence rates can be as high as 40% (2); hence close follow up with periodic physical examination is warranted. Metastatic epithelioid hemangioendothelioma of penis have been reported (2), but no established management policy due to paucity of cases. Interferon alpha and paclitaxel have been tried with some success (3).

Table 1: Different reported series of epithelioid hemangioendothelioma of penis

<table>
<thead>
<tr>
<th>Series</th>
<th>No of cases</th>
<th>Presentation(s)</th>
<th>Management(s)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Weiss and Enzinger</td>
<td>3</td>
<td>Solitary penile lesions with pain on erection</td>
<td>Local excision</td>
</tr>
<tr>
<td>Kamat et al</td>
<td>1</td>
<td>Solitary penile lesion</td>
<td>Surgery, interferon alpha and follow-up of 65 months without recurrence</td>
</tr>
<tr>
<td>Wen et al</td>
<td>1</td>
<td>Metastatic three chamber priapism</td>
<td>Partial excised, patient died</td>
</tr>
<tr>
<td>Gharajeh et al</td>
<td>1</td>
<td>Multifocal epithelioid hemangioendothelioma masquerading as superficial penile vein thrombosis</td>
<td>Surgically excised and did not recur after 1 year</td>
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</tbody>
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References

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