Actinomycosis of the Penile Shaft Coexisting with Fibrous Pseudotumor of the Testis

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Key Words: Penis; Testis; Actinomycosis; Fibrous tissue neoplasms

Actinomycosis of the penis is an uncommon malady; only five cases have been reported in the English literature.1-5 This rarity complicates pre-operative diagnosis. Moreover, this condition usually occurs at the corona of the uncircumcised penis and is associated with pilonidal sinus.1-5 Therefore, pre-operative diagnosis of actinomycosis of the penile shaft of a circumcised man is quite difficult.

Benign fibrous tumors of the testis and the paratesticular region are quite uncommon. They are classified as fibromas of gonadal stromal origin, fibromas of the testicular tunics, fibrosarcomatous lesions, fibrous pseudotumors and angiofibromatoses.6 These fibrous pseudotumors comprise about 6% of all paratesticular lesions.6,7

We report here a case of actinomycosis of the penile shaft not associated with the pilonidal sinus. This condition coexisted with fibrous pseudotumors of the testis.

CASE REPORT

A 37-year-old man presented with an ulcerated penile mass and slow-growing scrotal masses; these masses were first noted by the patient 10 years earlier. He had been circumcised 14 years ago. There was no inguinal lymphadenopathy and tests for sexually transmitted diseases were negative. One penile mass and eight scrotal masses were excised. The penile mass, with its healed surface ulceration, was located at the right side of the penile shaft (Fig. 1A). The penile mass measured 2.5 cm in diameter and the cut surface showed a yellow-brown color with central focal necrosis. The scrotal tumors were circumscribed, whorled, white masses 0.3-2.0 cm in diameters and were attached to the tunica vaginalis and tunica albuginea (Fig. 1B). Microscopically, the penile mass showed active inflammatory changes containing actinomyces displaying characteristic sulfur granules. Testicular masses were fibrous pseudotumors composed of bland spindle and stellate cells lying in dense collagenous stroma. Actinomycosis of the penis has been reported to occur at the corona of the uncircumcised penis associated with pilonidal sinus. The present case was not associated with pilonidal sinus and, unusually, displayed co-existence with fibrous pseudotumors of the testis.

Here, we present an uncommon case of the penile shaft actinomycosis with coexisting fibrous pseudotumors of the testis. A 37-year-old, circumcised man presented with one penile and eight scrotal masses. The penile mass having a healed surface ulceration was located at the right side of the penile shaft. It was relatively circumscribed without a fibrous capsule. The cut surface showed a yellow-brown color with central focal necrosis. The scrotal tumors were circumscribed, whorled, white masses 0.3-2.0 cm in diameters, and were attached to the tunica vaginalis and tunica albuginea. Microscopically, the penile mass showed active inflammatory changes containing actinomyces displaying characteristic sulfur granules. Testicular masses were fibrous pseudotumors composed of bland spindle and stellate cells lying in dense collagenous stroma. Actinomycosis of the penis has been reported to occur at the corona of the uncircumcised penis associated with pilonidal sinus. The present case was not associated with pilonidal sinus and, unusually, displayed co-existence with fibrous pseudotumors of the testis.

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Actinomycosis and Fibrous Pseudotumor

Infiltration mainly composed of neutrophils and histiocytes. The intermediate areas and periphery of the mass were composed of loose fibrous tissue with mononuclear inflammatory infiltration. However, there was no proliferation of spindle cells with dense collagenized stroma. The penile mass also showed sulfur granules consisting of a granular center surrounded by a radiating zone of eosinophilic, hyaline, club-shaped projections. The sulfur granules were surrounded by aggregates of neutrophils (Fig. 2A). Focally, foreign body giant cells were identified. Histochemically, Gram staining and Gomori’s methenamine silver stain revealed typical filamentous bacteria. The scrotal tumors were less cellular, with bland spindle and stellate cells lying in a dense collagenous stroma. Infiltration of inflammatory cells was present (Fig. 2B). The distribution of spindle cells in a collagenous stroma was homogeneous. Myxoid change and hemangiopericytoma-like vessels were not identified. Neither were cytologic atypia and mitosis. No infectious microorganisms such as Actinomyces were evident.

Immunohistochemically, the scrotal tumor was positive only for vimentin; stainings for cytokeratin, desmin, CD34, smooth muscle actin, S-100 protein, Bcl-2, and CD99 were negative. Postoperatively, the patient was treated with penicillin G (18 mU/day) intravenously for five days, followed by oral therapy with amoxicillin (1.5 g/day) for three months. Once treatment began, recovery was progressive. No recurrence was noted over four years later.

**DISCUSSION**

Genitourinary actinomycosis has been reported in the kidney, bladder and scrotum, but rarely in the penis.1–5 We found only six cases of actinomycosis of the penis in the Russian- and English-language literature (Entrez Pubmed, key words actinomycosis and penis). We reviewed all English language cases. Patients in these past and present cases of actinomycosis of the penis range
in age from 21-53 years. As shown in Table 1, four cases were associated with pilonidal sinus and the patients were uncircum-cised. There was no case where penile actinomycosis coexisted with fibrous pseudotumor of the testis. To our knowledge, ours is the first such case. However, there was no correlation between penile actinomycosis and the scrotal fibrous pseudotumors. There were no histologic features of fibrous pseudotumors such as pro-\[\textit{\text{in}}\]leration of spindle cells and dense collagenous stroma in the penile mass. Moreover, Actinomyces were absent in the scrotal masses. These two penile and scrotal lesions are, thus, likely, clinically separate, despite their co-existence.

A diagnosis of actinomycosis is typically made by the histologi-cal identification of actinomycytic granules or via anaerobic culture. Actinomyces are characterized by the formation of Gram-positive, branching filaments that are rimmed by Gram-negative, peripheral radiating clubs.\footnote{Val-Bernal et al.} Actinomyces generally have low virulence and they act as endogenous opportunists. The organisms grow in chronic ulcers, abscesses and sinuses, and they rarely produce any systemic disease.\footnote{1-5} In our case, there was no evidence of any pre-disposing factors such as pilonidal sinus, anorectal disease, previous trauma or surgery at this site, diabetes mellitus, steroid therapy, immunodeficiency or any symptomatic gynecological problem of the marriage partner. Although we could not find any predisposing factors for actinomycosis, we suggest that actinomycosis can occur in a circumcised patient. Physicians should be aware of this condition for patients who present with unusual or ulcerated lumps on the penis.

Because of the potential for actinomycosis to recur, prolonged antibiotic treatment is prudent; the exact duration of therapy depends on the site and severity of disease.\footnote{Penicillin is the generally accepted antibiotic of choice for both deep-seated and superficial forms of actinomycosis.} Penicillin is the generally accepted antibiotic of choice for both deep-seated and superficial forms of actinomycosis.\footnote{Surgical treatment may be mandatory when there are large collections of purulent and necrotic material.} Making the preoperative diagnosis was difficult in this case because of the coexistence of fibrous pseudotumors on the testis. Of particular note was the fact that the ulcerative lesion caused by the penile actinomycosis was suspected to be malignant; therefore, we performed surgery. After the diagnosis of actinomycosis, successful postoperative treatment with penicillin was conducted for three months. A prolonged observation period for the patient after treatment was necessary to detect possible recurrences.\footnote{We have followed the patient for four years to check for any relapse; he continues to be well.} Fibrous pseudotumors must be distinguished from other tumors of the testis and the paratesticular region such as fibromas of gonadal stromal origin, fibromas of the testicular tunics, fibrosarcomatous lesions, and angiomyofibroblastoma.\footnote{Fibrous pseudotumors are different from other paratesticular benign fibrous tumors. They usually present as multiple, well demarcated nodules composed of interlacing fascicles of bland looking fibrous tissue and chronic inflammatory cells with a dense collagenous stroma. Although most paratesticular masses containing a fibrous tissue component are benign, the differential diagnosis must include malignant spindle cell tumors, particularly malignancies such as fibrosarcoma and leiomyosarcoma. As in this case, the clinical presentation of fibrous pseudotumors may raise concern of an existing malignant process. However, necrosis, pleomorphism and mitotic figures that would suggest malignancy are not evident within fibrous pseudotumors.} In this case, there were no histologic findings of malignant tumors and we, therefore, excluded them in our diagnosis. Fibrous pseudotumors are also differentiated from solitary fibrous tumors by the existence of dense collagenized stroma. However, overall histologic findings are different. In this case, hemangiopericytoma-like vessels were not identified and the cells of scrotal lesions were negative for CD34. Therefore, we excluded solitary fibrous tumors in our diagnosis. Fibrous pseudotumors are regarded as reactive inflammatory lesions. Many fibrous pseudotumors are composed of interlacing fascicles of fibrous tissue and chronic inflammatory cells, and have peripheral neovascularity, suggesting a traumatic or reactive histogenesis.\footnote{Therefore, mass excision is sufficient for treating fibrous pseudotumors of the testis.} In this case, mass excision was also sufficient for treating the patient. Familiarity with fibrous pseudotumors may prevent such unnecessary and radical treatment as orchietomy.

Table 1. Clinicopathologic characteristics of this case of actinomycosis of the penis and the five previously reported cases

<table>
<thead>
<tr>
<th>Authors</th>
<th>Ref</th>
<th>Age</th>
<th>Site</th>
<th>Symptom</th>
<th>Status of circumcision</th>
<th>Associated disease</th>
</tr>
</thead>
<tbody>
<tr>
<td>Chikkamuniyappa et al.</td>
<td>1</td>
<td>22</td>
<td>Foreskin</td>
<td>Cystic nodule</td>
<td>Uncircumcised</td>
<td>Pilonidal sinus</td>
</tr>
<tr>
<td>McParland et al.</td>
<td>2</td>
<td>31</td>
<td>Penis</td>
<td>Mass on the penis</td>
<td>Unknown</td>
<td>Absence</td>
</tr>
<tr>
<td>Rashid et al.</td>
<td>3</td>
<td>21</td>
<td>Corona</td>
<td>Rapidly growing lump</td>
<td>Uncircumcised</td>
<td>Pilonidal sinus</td>
</tr>
<tr>
<td>Sahayar et al.</td>
<td>4</td>
<td>53</td>
<td>Corona</td>
<td>Painful swelling</td>
<td>Uncircumcised</td>
<td>Pilonidal sinus</td>
</tr>
<tr>
<td>Val-Bernal et al.</td>
<td>5</td>
<td>25</td>
<td>Corona</td>
<td>Recurrent abscess</td>
<td>Uncircumcised</td>
<td>Pilonidal sinus</td>
</tr>
<tr>
<td>Present case</td>
<td>37</td>
<td></td>
<td>Penile shaft</td>
<td>Mass on the penis</td>
<td>Circumcised</td>
<td>Fibrous pseudotumor of the testis</td>
</tr>
</tbody>
</table>

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REFERENCES


