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Case report

Gummatous penile syphilis

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ABSTRACT

Syphilitic gumma involving the penis is a rare manifestation of tertiary syphilis. Only seventeen cases have been reported in the literature. It can mimic other diagnoses such as penile carcinoma. We report a case of a 56 year old male that had been sexually abstinent for over 10 years and presenting with a 4 cm painful penile lesion with clinically palpable bilateral inguinal nodes with no prior history of sexually transmitted diseases (STDs). A positron emission tomography-computed tomography scan identified the penile mass as being hypermetabolic and suspicious for penile carcinoma. Several inguinal and pelvic lymph nodes were also found to be suspicious for penile carcinoma. A penile biopsy was proposed and declined by the patient as he opted for a partial penectomy. The surgery was performed for diagnostic and palliative purposes. Histopathological studies revealed the presence of polymorphous, granulomatous, epitheloid inflammatory infiltrate with giant cells. Additional microbiologic testing confirmed the diagnosis of tertiary syphilis, presenting as gummatous syphillis associated with neurosyphilis. The patient was treated with intravenous penicillin and had adequate clinical clinical and serologic 12 months following treatment. Gummatous syphillis is a rare entity, but should be considered in the differential diagnosis of a penile lesion. To rule out this possibility, a biopsy should always be performed prior to invasive penis surgery.

Introduction

Gumma involving the genitalia are rare, occurring in the context of tertiary syphilis and represent a significant diagnostic challenge. To our knowledge, only 17 cases have been reported since the early 20th century, the latest dating back to 1977 [1,2]. These lesions generally manifest in the bones, skin or mucocutaneous tissues as an infiltrative mass [3]. Thus, one could easily overlook a gumma as a differential diagnosis of penile lesion. We present a case of a syphilitic gumma of the penis and the relevant literature associated.

Case presentation

A 56 year old retired Navy sailor, known with a remote history of gastric carcinoma, was referred to our clinic by the medical oncology team for a slowly growing penile mass, suspicious of penile carcinoma. The lesion was first noted by the patient 3 months before presentation. Over this period, the patient experienced increasing penile pain requiring use of narcotics and complained of fatigue, anorexia and progressive weight lost as well as increasing obstructive lower urinary tract symptoms. Additionally, the patient suffers from erectile dysfunction and had been sexually abstinent for over 10 years. He had no prior history of sexually transmitted diseases (STDs).

On physical examination, an indurated ulcero-necrotic penile mass of 4 cm involving the glans and the distal penile shaft was noted, along with a partial phimosis. The groin examination revealed bilateral mobile painless lymph nodes of less than 2 cm. A positron emission tomography-computed tomography (PET-CT) scan demonstrated the presence of a 4 cm hypermetabolic penile mass along with multiple hypermetabolic bilateral inguinal and pelvic lymph nodes of less than 2 cm (Figs. 1 and 2). These findings were suspicious of penile carcinoma.

Based on the strong suspicion of penile carcinoma, a penile biopsy was first proposed to the patient, but declined. Given the severe intractable penile pain and considering that the patient had been abstinent for over 10 years, the decision was taken to perform **a** partial penectomy for palliative and diagnostic purposes (Fig. 3). Pathological assessment of the specimen revealed the presence of

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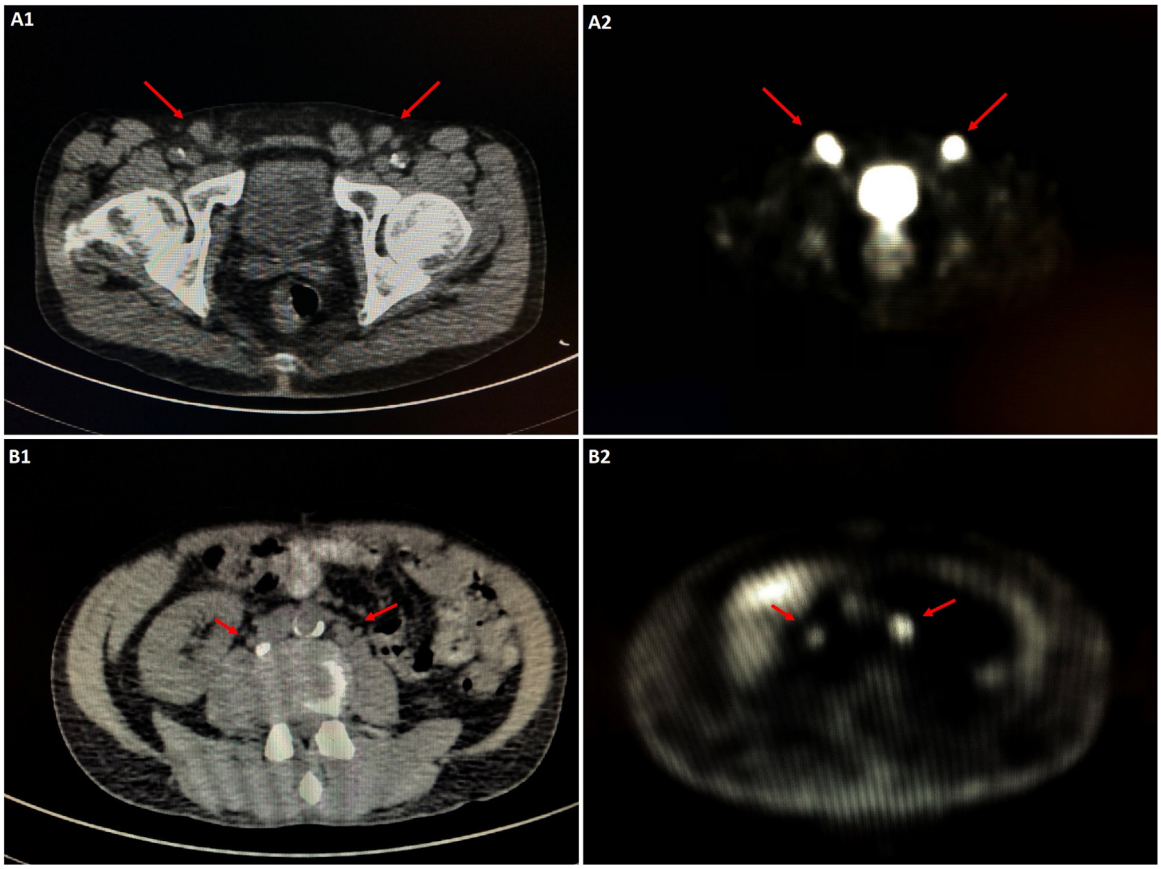

Fig. 1. PET scan showing hypermetabolic bilateral inguinal (A) and retroperitoneal lymphadenopathies (B).

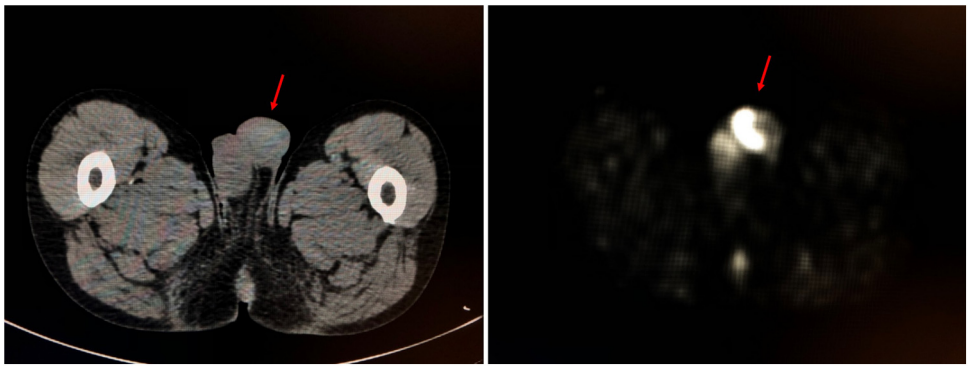

Fig. 2. PET scan showing hypermetabolic penile mass.

polymorphous, granulomatous, epitheloid inflammatory infiltrate with giant cells, associated with a focal abscess and a prominent vascular involvement (Figs. 4–6). No evidence of malignancy was identified. Specific tests to identify an infectious cause were negative, including histological (silver stain) and immunohistochemical search for *Treponema* pallidum.

One month after surgery, the patient developed neurological symptoms including visual disturbances, migraine and weakness. A nontreponemal rapid plasma regain (RPR) test (Macro-Vue RPR Card, Becton, Dickinson and Company, Franklin Lakes, NJ) revealed high titers (1:256). On lumbar puncture (LP), cerebrospinal fluid (CSF) analysis revealed a WBC count of 80×10^6/L (81% mononucleated cells, 11% neutrophils), a normal glucose level of 3.6 mmol/L, and elevated protein (0.69 g/L). A CSF RPR test was positive at 1:4. HIV testing was negative. On follow-up questionnaire, the patient admitted having several sexual intercourses with European and Caribbean sex workers in the 80's. The final diagnosis was tertiary syphilis with gumma and neurosyphilis. The patient was treated with intravenous penicillin G for 14 days, followed by a single dose of intramuscular benzathine penicillin. He is presently doing well with no residual symptoms. Follow-up nontreponemal testing showed adequate response and a PET-CT performed 9 months after initiation of therapy showed complete regression of lymphadenopathies.

Discussion

Currently there is an increase in the number of cases of infectious syphilis [4–6]. The previous outbreak was seen in the late 1980s, which means that tertiary syphilis cases being

Fig. 3. Macroscopic specimen. Note the yellow necrosis (red arrow) visible at the superior portion of the lesion. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

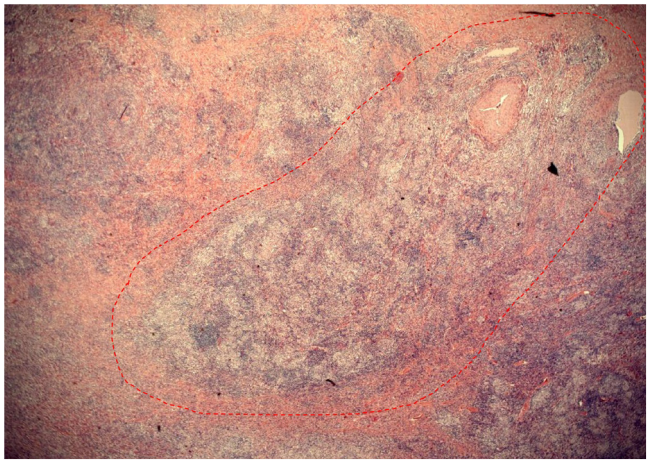

Fig. 4. Granuloma (encircled). (x20).

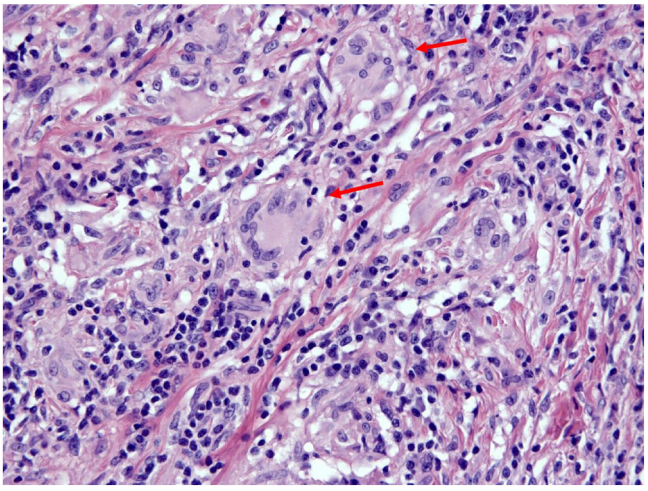

Fig. 5. Granuloma with giant cells (red arrows). (x400). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

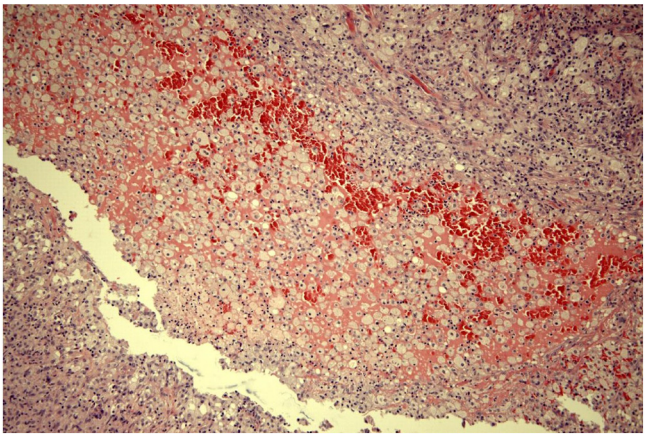

Fig. 6. Peri-abcess macrophages granuloma.

encountered now probably reflects that last outbreak. Gummatous disease is a rare manifestation of tertiary syphilis in the post-penicillin era. It can be misdiagnosed as cancer, as the lesions are often characterized by their local destructiveness, masslike effects, or both [7]. They are generally slowly growing, painless and they can manifest early (less than 1 year from exposure), but have been reported up to 46 years after secondary syphilis diagnosis [8].

Microscopically, gummas are composed of a central zone of coagulative necrosis surrounded by lymphocytes, plasma cells, epitheloid cells and occasionally giant cells. Spirochetes are only rarely identified in the lesion in contrast to the chancre seen in primary syphilis, where treponemas are visible on the surface of the ulcer with silver stains [9]. Gummatous syphilis is characterized by rapid improvement following penicillin therapy and previous gumma are usually replaced by scar tissue [10].

The most common sites for gummas are the skin, mucocutaneous tissue, bones and liver. Other uncommon sites such as central nervous system have also been described [11]. Gumma involving the genitalia is an even rarer presentation with only 17 cases having been reported in the literature since the 1950s [1,2]. The last documented case was reported in 1977. All seventeen cases were diagnosed based on physical examination and on serologic or histologic studies. All cases had complete resolution following antibacterial therapy.

In this case, the gumma was initially misdiagnosed as penile cancer. Contributing factors included the lack of clinical suspicion based on the sexually abstinent state of the patient, the appearance and painful presentation of the penile lesion and most importantly, the fact that clinicians, nowadays, are rarely exposed to gumma and tertiary syphilis. The physical examination and the PET-CT scan

findings were also highly suggestive of metastatic penile cancer. Nevertheless, a penile biopsy prior to the intervention would have likely shown granulomas with epithelioid and giant cells, obliterative endarteritis and necrosis, the characteristic gummatous form which would have likely prompted further investigations. When evaluating penile lesion, numerous differential diagnosis should be evoked, including neoplastic, infectious and inflammatory. Penile cancer can generally be diagnosed based on the history and examination alone. Obtaining pathologic samples is paramount in the management of penile cancer prior to initiating therapy [12]. On the other hand, biopsy of the lesion prior to surgery is not mandatory when the diagnosis is obvious according to guidelines from the European Urological Association [13]. However, a biopsy is recommended when there is doubt about the exact nature of the lesion or when topical, laser or radiotherapy treatment is considered. Overall, biopsies proved to differ from final grade in 30% of cases and failed to detect cancer in approximately 3.5% of cases [14].

Conclusions

Syphilitic gumma of the penis is a rare entity. Nevertheless, it should still be considered in the differential diagnosis of penile lesion, even in the absence of recent intercourse as tertiary syphilis will often present years to decades after exposure. Given the cosmetic and functional morbidity associated with a partial penectomy, a biopsy should always be considered prior to surgery.

Informed consent

Obtained

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