

LETTER TO THE EDITOR

Penile metastasis of osteosarcoma: a rare case report

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Dear Editor,

Metastasis to the penis is rare, with no more than 500 cases reported to date.^{1–4} Primary tumours of sarcomatous origin are extremely rare, and no case of osteosarcoma has been described in the literature. Here, we present a case of penile metastasis from osteosarcoma of the ischial tuberosity and discuss its possible metastatic mechanisms and clinical implications.

A 42-year-old man, who had a known diagnosis of osteosarcoma of the ischial tuberosity on the right side, presented with a 2-week history of painful nodules in the glans penis in October 2011. He had already been diagnosed with osteosarcoma of the right ischial tuberosity with pulmonary metastasis in the left lower lobe 2 months before presentation (**Figure 1a**). Six weeks prior, he had undergone resection of the tumour of the ischial tuberosity. Two weeks prior, he had noticed the onset of two painful nodules in the glans penis. His condition was initially considered to be Peyronie's disease or an infectious disease. Oral antibiotics were prescribed, but the treatment was ineffective. Upon physical examination, there were two erythematous nodules in the glans penis, each measuring 5×5 mm (**Figure 1b**). There was no discharge, ulceration or necrosis on the overlying skin of the glans, and no priapism phenomenon was observed. The superficial inguinal lymph nodes were not obviously enlarged. At this time, a computed tomography (CT) scan of the pelvis was performed, but yielded no specific findings for the penis, and a skeleton emission CT scan was normal. On suspicion of malignancy, a biopsy of the nodules was performed, and histological examination was consistent with metastatic osteosarcoma (**Figure 1c and 1d**). Immunohistochemistry showed the tumour to be CD3⁺, CD20⁺, CD117⁺, CK⁺, CgA⁺, LCA⁺, SY⁺, inhibin a⁺ and vimentin⁺. The patient and his family were counselled extensively about the extent of the patient's disease and about treatment options, but he refused any further therapies. He died 3 months later.

This case was presented for several reasons. First, this case was extremely rare. Primary tumours that most commonly metastasize to the penis are those of the bladder and prostate, followed by rectosigmoid tumours. Other primary tumours include those of the kidney, pancreas, liver, nasopharynx and lung; malignant melanoma; and Burkitt's lymphoma.^{1–7} Primary tumours of sarcomatous origin are extremely rare, and only one case of chondrosarcoma⁸ and one case of penile primary osteosarcoma⁹ have been reported. To the best of our knowledge, this is the first case of penile metastasis from osteosarcoma to date.

Second, this case might present a rare metastatic mechanism. The mechanisms of penile metastasis have been under discussion, and the following possible mechanisms have been proposed: metastasis *via* the retrograde venous route, the retrograde lymphatic route, direct extension and arterial spread.⁷ The retrograde venous route is the most likely mechanism of metastasis because there is rich communication between the dorsal penile venous system and the pelvic organs. This mechanism can explain most secondary tumours arising from the prostate, bladder and rectosigmoid. Metastasis *via* the retrograde lymphatic route is thought to occur in a fashion similar to spread *via* the retrograde venous route. Direct extension is generally observed in rigorously invasive tumours originating from sites close to the penis. Arterial spread, however, is considered to be uncommon, and no case has been presented to date.

In the current case, we consider the most likely mechanism to be arterial spread, for the following reasons: (i) general pattern of sarcomatous metastasis. The dissemination of osteosarcoma frequently occurs through haematogenous spread instead of lymphatic spread;¹⁰ (ii) characteristics of symptoms. In the retrograde route or direct extension, metastatic lesions are commonly localized to the proximal penile shaft,^{6,7} and occlusion of the draining veins may cause priapism. However, in the present case, lesions were found in the glans of the penis, and priapism was absent throughout the disease process; and (iii) origin of tumour emboli. The tumour originated from the ischial tuberosity and resided in the pelvic cavity. The arteries that supply the penis could have been invaded, in which case the tumour emboli would be delivered into the penis. Thus, arterial spread seems to be a reasonable explanation in this case.

Third, this case emphasized the importance of biopsy for unclassified penile nodules. The most common symptom of penile metastasis is priapism, and other clinical manifestations include penile masses or nodules, ulceration, obstructive or irritative urinary symptoms and haematuria.^{1–4,7} In this case, the only symptom was penile nodules, which were misdiagnosed as Peyronie's disease at first. Although penile metastasis and Peyronie's disease have a similar manifestation of painful penile nodules, certain differences are noted: (i) penile metastasis has a tumour history, whereas Peyronie's disease does not; (ii) penile metastasis has a rapid progression, whereas Peyronie's disease has a relatively slower progression; (iii) the plaque associated with Peyronie's disease is mainly confined to the corpus cavernosum, and penile metastasis may involve the glans penis, as shown in this case. Penile metastasis is easily misdiagnosed because

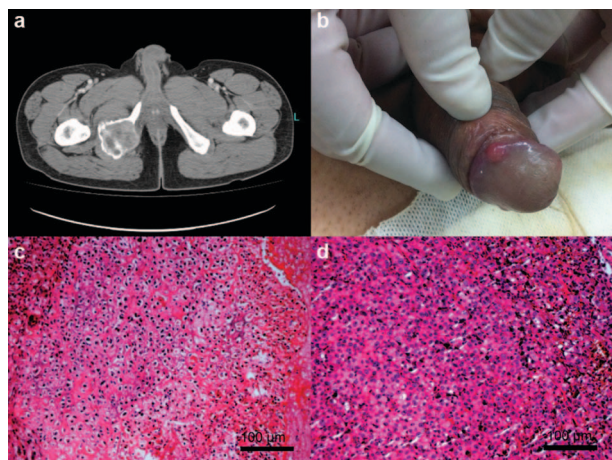


Figure 1 (a) CT scan of the primary osteosarcoma from the ischial tuberosity. (b) Gross examination of the metastatic nodules on the glans penis. (c) Microscopic examination of the primary tumour. Haematoxylin-eosin stain, scale bar=100 µm. (d) Microscopic examination of the penile metastases. Haematoxylin-eosin stain, scale bar=100 µm. CT, computed tomography.

it is rare, and the only method for establishing a correct diagnosis is a biopsy of the nodules with pathological examination. Thus, we propose that biopsy should be considered if unusual, rapidly progressing penile nodules occur in tumour patients.

Fourth, the association between penile metastasis from osteosarcoma and poor prognosis was also shown in this case. The expected survival time of patients without any treatment and with primary tumours originating from the urogenital tract is approximately 6

months.^{6,7} In the current case, the survival time was only 3 months, suggesting more aggressive behaviour for metastatic osteosarcoma.

AUTHOR CONTRIBUTIONS

NL cared for the patient, collected clinical information and drafted the manuscript. LBM and GLH revised the paper and participated in critical discussion. All authors read and approved the final manuscript.

COMPETING FINANCIAL INTERESTS

The authors declare no competing financial interests.

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