

CASE REPORT

Penile Mondor's disease in a patient treated with radical chemoradiation for anal cancer

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Abstract

Penile Mondor's disease is a rare condition characterized by sclerosing thrombophlebitis of the superficial dorsal penile vein. Usually its causes are benign, but it is also evident in cancer patients. We report the case of a 62-year-old man with a cT4 anal cancer (infiltration of corpora spongiosa and penile bulb), associated with extensive loco-regional lymphadenopathy, who developed painful lumps in the midline of the anterior penile surface while receiving radical chemoradiotherapy. Physical examination revealed two palpable cord-like swellings located 2 cm from the pubic symphysis. Color Doppler ultrasound established the diagnosis of Mondor's disease. The patient was successfully managed with non-steroidal anti-inflammatory drugs. The causative factors were pelvic malignancy and radiotherapy. The diagnosis was challenging since Mondor's disease is a rare condition and the differential diagnosis included malignancy progression. This is the first case report describing penile Mondor's disease in a patient with anal cancer under chemoradiotherapy.

INTRODUCTION

Phlebitis of the dorsal veins of the penis in a patient with generalized phlebitis was first described by Braun Falco in 1858 [1]. However this clinical entity took its name from Mondor in 1939, who described thrombosis of the superficial thoracoepigastric veins in female patients [2]. Its incidence in patients with sexually transmitted diseases (STD) was reported to be 1.3%, but it is strongly believed that in reality it is higher, as affected patients do not seek medical help mainly due to concern that this could represent a sexually transmitted disease and the possible associated social stigma [2, 3]. The clinical picture of Mondor's penile disease is typical and in the event of its diagnosis the possibility of an underlying malignancy should be investigated.

CASE REPORT

A 62-year-old man was diagnosed with a locally advanced cT4 anal cancer (squamous cell carcinoma) involving adjacent anatomical structures (such as the prostate, posterior part of the membranous urethra, penile bulb, bulbourethral gland and the corpora spongiosa) and bilateral inguinal lymph nodes. Moreover, staging MRI of pelvis/anus reported pathologic lymph nodes measuring up to 1 cm, located anterior to the pubic symphysis (Fig. 1). Baseline physical examination of the patient's genitalia showed no abnormality. The patient was managed with radical radiotherapy (IMRT technique) combined with chemotherapy (mitomycin: 10 mg/m² on Day 1 and 5-FU: 1000 mg/m²/24 h on Days 1–4). Chemotherapy was administered during the first and last weeks of radiotherapy.

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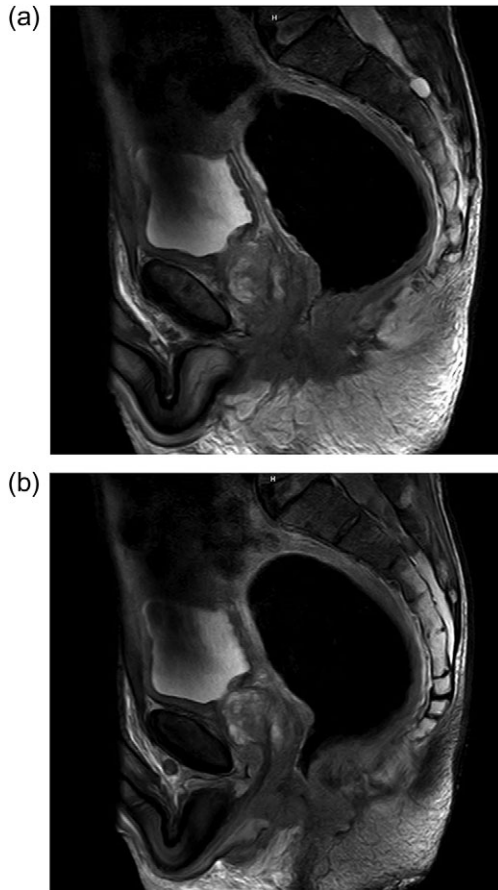


Figure 1: T2 saggital MRI images. (a) The anal carcinoma infiltrates the inferior aspect of the prostate, the penile bulb, the regional corpora spongiosa and the posterior area of the membranous urethra. (b) Pathologic lymph node located anterior to the pubic symphysis.

With respect to the radiation treatment schedule, the patient received 45 Gy to the clinical target volume and 54 Gy to the gross tumor volume, which included the primary tumor, involved anatomical structures and pathological lymph nodes. Radiotherapy was delivered in 25 fractions. It is essential to note that no particular set up for the penis was used, with the patient being instructed to place his penis in a downward relaxed position before each radiotherapy session. No strict dose constraint was used for the genitalia, since these were involved by the tumor and pathological lymph nodes were diagnosed in the medial aspect of the inguinal regions and anteriorly to the pubic symphysis. The maximum dose delivered in the area of the development of Mondor's disease was 35Gy.

During his admission for the second chemotherapy cycle (delivered radiotherapy dose 43.2 Gy) the patient reported painful lumps in the midline of the anterior penile surface, 2 cm away from the pubic symphysis. There was no other alarming symptom like hematuria, itching, dysuria or fever. The patient was under treatment with an angiotensin II receptor antagonist for high blood pressure and denied any history of recent trauma, sexual activity, masturbation or the use of constrictor devices. He had never experienced this before and he denied a history of STD.

Physical examination revealed two palpable rope-like painful lesions, measuring about 5 mm each, in the aforementioned

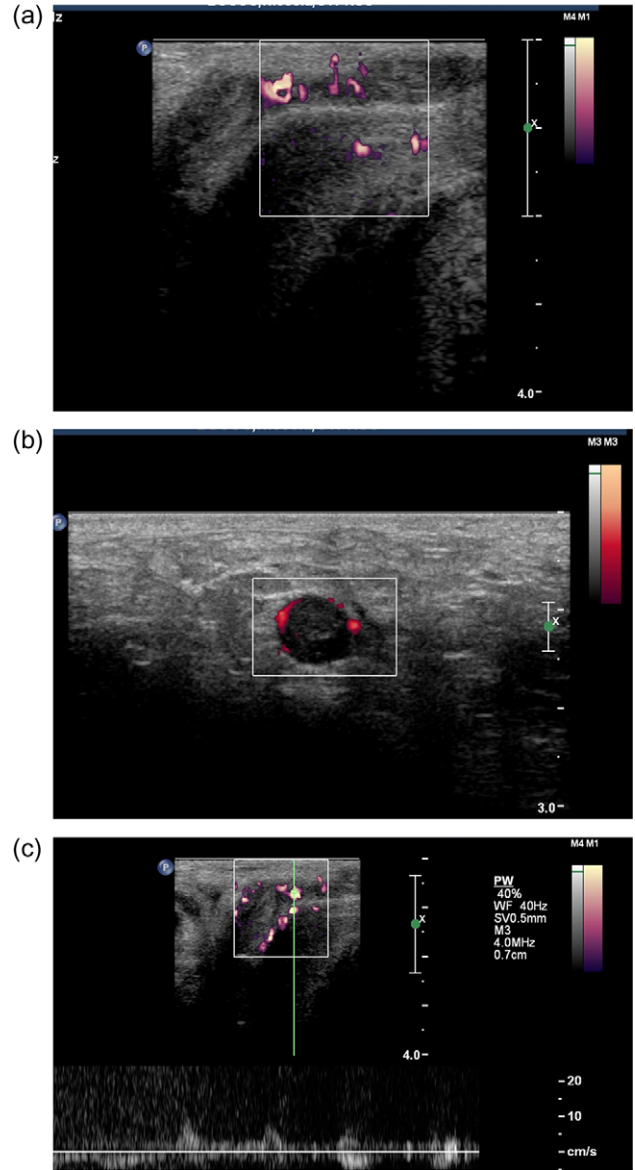


Figure 2: Penis ultrasound. Gray scale mode with power Doppler. The deep dorsal vein is completely occluded due to thrombosis. Collateral circulation is noted at the periphery of the vein. (a) Longitudinal visualization, (b) Axial section and (c) Spectral analysis of the blood flow in collateral vessels.

area. What was worrying and challenging was the possibility of disease progression since in the medial aspect of the groin area and anterior to the pubic symphysis pathologic lymph nodes were diagnosed, only a few cm away. Moreover, the penile bulb and corpora spongiosa were infiltrated by the tumor. The differential diagnosis also included thrombosis.

Routine blood tests were normal, whereas Doppler ultrasound examination showed a non-compressible portion of superficial dorsal vein, as well as the absence of venous flow signals (Fig. 2). The surrounding soft tissues were hyperechoic due to associated inflammation. The patient was managed with non-steroidal anti-inflammatory drugs (NSAIDs) (ibuprofen 400 mg twice a daily) and was instructed to abstain from any sexual activity until symptoms resolved. Four weeks after the diagnosis of Mondor's disease physical examination was normal and the patient reported resolution of symptoms.

The patient has currently completed 1 year of follow up and has a complete clinical and radiological response.

DISCUSSION

Mondor's disease is more frequently diagnosed in the thoracoepigastric region and usually affects middle aged women. Less commonly the penis, upper arms, arm pits and groins are affected [4]. Thrombosis of the dorsal vein of the penis is a rare disorder that affects males in the age range between 20 and 70 years [4]. The pathophysiology of the disease is not well known and in many cases the etiology remains uncertain. Several pathogenic factors have been proposed, such as Virchow's triad vessel wall damage, vascular stasis and coagulation disorders [2, 5]. Well known causes include prolonged vigorous sexual activity and STD [3]. Postsurgical vascular stasis, local or distant inflammatory processes, excessive exercise, pelvic tumors and injection of intravenous drugs have been also described as causative factors [2, 5]. Among malignancies the co-existence of breast cancer and Mondor's disease has been described most in the literature. The incidence in such patients reaches 12.7% [6]. Therefore in the event of Mondor's disease diagnosis the possibility of malignancy should be investigated [6]. In the presented case both the diagnosis of pelvic malignancy and radiotherapy are predisposing factors for the development of thrombosis, since radiotherapy may cause endothelial damage [7].

Even though thrombosis is frequently diagnosed in cancer patients and has been associated with antineoplastic treatments, to the best of our knowledge this is the first case report describing the diagnosis of Mondor's disease in a patient with anal carcinoma receiving radical radio-chemotherapy. Medical history and thorough physical examination set the clinical diagnosis of penile Mondor's disease. The thrombosed dorsal vein becomes thickened and adherent to the neighboring tissues giving the impression of a swollen cord. Additionally associated edema may be present in the affected region [5]. Sclerosing lymphagitis and Peyronies disease should be excluded since they are included in the differential diagnosis [5]. Moreover, if clinical uncertainty remains, color Doppler ultrasound can establish Mondor's disease. Ultrasonography shows a non-compressible distended vein [2]. Last but not least, as far as the primary workup is concerned, coagulation should be always evaluated in routine laboratory tests, as the underlying cause may be related to coagulation disorders [5].

Usually penile Mondor's disease is a self-limiting process and resolves within 4–6 weeks after diagnosis. Further thrombosis is not usually observed and anticoagulation treatment is not recommended [5]. The overall management is limited to best supportive care, including pain management, warm compresses (unless presence of dermatitis) and instructions for abstinence from sexual activity. NSAIDs (ibuprofen 400 mg or diclofenac 50 mg taken twice a daily) are commonly prescribed because of their double activity as pain killers and anti-inflammatory drugs [2]. For pain management in patients with refractory pain, opioid analgesics can be used in combination with NSAIDs or as a monotherapy. In cases with bleeding

carcinomas NSAIDs and anticoagulation should be avoided. If there is suspicion of STD or there are signs of infection, indicated treatment should be administered [2, 3]. If symptoms last for more than 6 weeks, or the patient reports persistent pain, vein resection could be considered [2, 8].

In conclusion, the diagnosis of Mondor's disease in this patient was challenging as this is a rare diagnosis. Additionally, the differential diagnosis included disease progression, with the management of the two being totally different. Therefore, appropriate clinical suspicion and investigations led to the correct diagnosis and management.

CONFLICT OF INTEREST STATEMENT

None declared.

FUNDING

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ETHICAL APPROVAL

Ethical approval was given from B.O.C. Oncology Center for publication. The patient is not identifiable in any of the images provided.

CONSENT

The patient has consented to publication of the case.

REFERENCES

- Bird V, Kasnokutsky S, Zhou H, Jarrahy R, Khan SA. Traumatic thrombophlebitis of the superficial dorsal vein of the penis: an occupational hazard. *Am J Emerg Med* 1997;15: 67–9.
- Wash JC, Poimboeuf S, Garvin DS. A common presentation to an uncommon disease. Penile Mondor's disease: a case report and a literature review. *Int Med Case Rep J* 2014;7:155–7.
- Kumar B, Narang T, Radotra BD, Gupta S. Mondor's disease of the penis: a forgotten disease. *Sex Transm Infect* 2005;81: 480–2.
- Fietta P, Manganelli P. Mondor's diseases. Spectrum of the clinical and pathological features. *Minerva Med* 2002;93: 453–6.
- Nazir SS, Khan M. Thrombosis of the dorsal vein of the penis (Mondor's disease): a case report and review of the literature. *Indian J Urol* 2010;26:341–433.
- Catania S, Zurrada S, Veronesi P, Galiberti V, Bono A, Pluchinotta A. Mondor's disease and breast cancer. *Cancer* 1992;69:2267–70.
- Blann AD, Dunmore S. Arterial and venous thrombosis in cancer patients. *Cardiol Res Pract* 2011;2011:394740. doi: 10. 4061/2011/394740.
- Al-Mwalad M, Loertzer H, Wicht A, Fornara P. Subcutaneous penile vein thrombosis (penile Mondor's disease): pathogenesis, diagnosis, and therapy. *Urology* 2006;67:586–8.