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Mondor's disease of the penis due to asymptomatic infective prostatitis provoking episodes of secondary sclerotizing lymphangitis

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Abstract

Mondor's disease of the penis is an under-reported condition caused by superficial thrombophlebitis of the dorsal vein or thrombosis of the deep venous network of the penis. This is a benign, self-limiting condition, characterized by a sudden, indurated swelling of the aforementioned veins. The possible causes comprise traumatism, neoplasms, excessive sexual activity, or coagulation inherited deficit. An accompanied lymphangitis is discussed, eventually as a distinct variety. The differential diagnosis must be established by the means of ultrasound, which is the imaging diagnostic technique of choice. We describe the case of Mondor's sclerotizing lymphangitis secondary to recurrent episodes of genitourinary infection previously diagnosed as recurrent superficial Mondor thrombophlebitis of the penis.

Case Report

A 44-year-old Italian Caucasian heterosexual man presented to our Vascular Disease Clinic concerned with a 5-6 days of mildly painful inflammatory indurated structures at the penile dorsum and erection discomfort. The patient reported a several years history of recurrent Mondor's disease of the penis. Previous medical history included hypothyroidism, gastroesophageal reflux disease, an episode of prostatitis with *Escherichia coli* urinary tract infection in July 2020, but denied any family or personal history of deep venous thromboembolism, coagulation disorders, and periods of immobility or surgery. The patient was generally physically well, not overweight, and did not smoke. On examination, the patient had no cord-like lesion palpable within the dorsal vein of the penis, but at the base of the penile shaft was poorly compressible, minimally tender with no oedema or cellulitis evident. High-resolution ultrasonography examination was otherwise unremarkable for signs of superficial thrombophlebitis or thrombosis neither of the dorsal vein of the penis, nor of the deep cavernous veins. On the contrary, the presence of an enlarged lymph node at the basis of the penis, encircled by several micro hyperechoic spots suggested possible recurrent episodes of lymphangitis (Figure 1). This diagnostic interpretation was further reinforced by the images of calcificated lymphadenopathy bilaterally in the groin region (Figure 2). For such a reason we planned a full microbiologic genitourinary screen revealing in the seminal fluid the significant presence of *Citrobacter koseri.* The latter represented the microbial agent of an asymptomatic prostate colonization causing recurrent episodes of urogenital bacterial infection which clinically was manifested with synchronized episodes of acute lymphangitis. They were previously interpreted and accordingly treated as Mondor's thrombophlebitis, with scarce results. The patient was finally treated with specific antibiotics and nonsteroidal anti-inflammatory drugs for about 4 weeks (bactrim 800 mg + 160 mg for 4 weeks and metronidazole 250 mg for 2 weeks, followed by humatin 250 mg for 2 weeks). At three months follow-up no more symptoms nor erectile disfunction has been recorded.

Discussion

Mondor's disease (MD) was first reported in the early 1850s by Henri Mondor, a French surgeon, who described it in detail only in 1939.[1] Although no formal classification has yet been established, MD on the anterolateral thoracoabdominal wall is generally recognized as original MD, while similar abnormalities on other sites, such as the penis and axilla, are recognized as variants of MD.[2] The former is called penile MD (first reported by Helm *et al*., in 1958[3]) and the latter is called axillary web syndrome (first reported as a complication of axillary surgery by Moskovitz *et al*. in 2001.[4] Almost all cases of MD are reported to be thrombophlebitis of the superficial vein, although some are reported to be lymphangitis and/or a combination of both the etiologies.[5] MD occurs with a palpable, subcutaneous, painless or tender, cord-like induration, beneath the skin on the chest wall along front axillary line.[2] In most cases the superficial thickening corresponds to the route of the thoraco-epigastric vein and is caused by a superficial thrombophlebitis.[6] The thoraco-epigastric vein connects the superficial epigastric vein and the lateral thoracic vein, and therefore drains into both the superior vena cava *via* the axillary vein and the inferior vena cava via the femoral vein.[6] According to the Heijblom *et al.* investigation, such a vein, running at a depth of 2 to 3 mm, can be perceived by the human retina as a bluish discoloration of the chest wall.[7]

Penile MD (PMD) is a rare entity, with reported incidence of 1.39%[7] in men 20-40 years of age.[5] There seems to be an underestimated incidence because it is a self-limiting process with recanalization of the vein after one to two months, rare doctor's consultations, and often incorrect diagnoses.[8,9] The etiology is still controversial. Causes of the penile Mondor's disease are multifaceted: excessive sexual activity or prolonged sexual abstinence, priapism or penile strangulation due to a tumor or distended bladder, penile trauma or injections, urogenital infections, pelvic surgery and prostate biopsy, hematologic diseases, thrombophilia, and paraneoplastic syndrome. Endothelial dysfunction, stasis of blood flow and hypercoagulability (due to excessive inflammation or platelet activation), also known as Virchow's triad may predispose patients to thrombotic disease, both in the venous and arterial circulations.[2] A history of deep venous thrombosis of the leg, migratory phlebitis associated with cancer, and surgical repair of an inguinal hernia can be considered risk factors.[10] In the literature is also reported a case of penile Mondor's disease, that has occurred after a long-haul flight, which required to maintain a sitting position for a long time.[11] The application of recently

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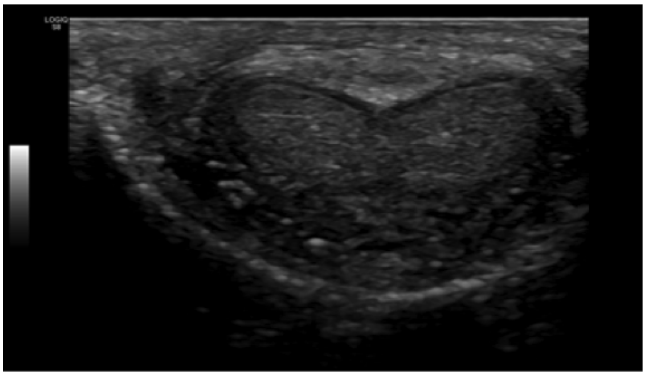

Figure 1. Absence of superficial and/or deep cavernous vein thrombosis related imaging of the penis. A lymph node is enlarged at the basis of the penis, and encircled by several micro hyperechoic spots. The latter could represent the traces of recurrent episodes of lymphangitis.

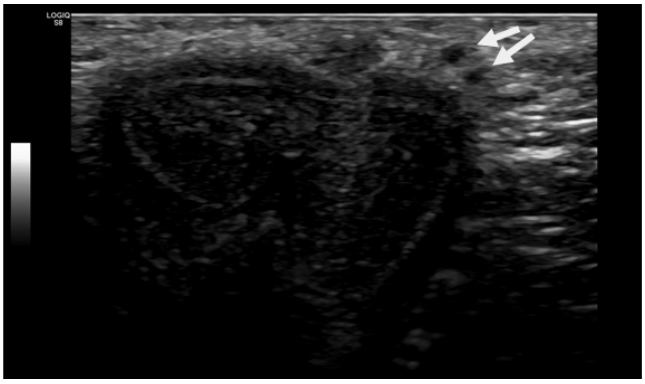

Figure 2. Calcificated inguinal lymph nodes.

developed immunohistochemical markers revealed that almost all Mondor's diseases appeared to be thrombophlebitis of the superficial vein, while some cases occurred due to lymphangitis.[12] The combination of polyclonal antibody against human lymphatic vessel endothelial hyaluronan receptor-1 (LYVE-1, which is a marker for the lymph vessel) and von Willebrand factor antibody (a conventional marker of the blood vessels) can clearly distinguish between these two vessels. In addition, sclerotizing lymphangitis of the penis, which is clinically identical to PMD, was found to be thrombophlebitis in most cases using monoclonal antibodies against CD31 and CD34.[8] Although it has a short, self-limiting benign course, patients experience a firm, string-like induration in the coronal sulcus of the penis and along the penile superficial dorsal vein, which is often painful and anxiety-provoking. The vein may appear to be swollen and erythematous. The patient will report having a significant amount of pain, which can be either episodic or constant.[10] The diagnosis is usually based on clinical findings, but Doppler ultrasound can be useful in cases of diagnostic uncertainty, where it may reveal a non-compressible distended vein. The patient do not require further investigation unless clinically indicated.[9] Spontaneous resolution following venous recanalization occurs within a mean of 4 weeks, but can take up to 8 weeks.[9] Non-steroidal and anti-inflammatory drugs may be used for symptomatic relief, although they do not affect resolution rate.[13] Anticoagulants are not necessary and may cause excess blood leakage if a ruptured corpus cavernosum is incorrectly diagnosed as dorsal vein thrombophlebitis.

Conclusions

We herein describe a rare case of sclerotizing lymphangitis secondary to genitourinary bacterial colonization by *C. koseri,* as a consequence of a recurrent prostatitis. This contributes to define secondary sclerotizing lymphangitis as a variant of penile Mondor's disease. We may accordingly conclude that ultrasound is able to discriminate between sclerotizing lymphangitis and thrombophlebitic varieties of the Mondor's disease of the penis, suggesting therefore the proper treatment.

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