

Scrotal Swelling and Penile Fistula in a Patient with Crohn's Disease

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ABSTRACT

Crohn's disease often is detected in adolescents with nonspecific gastrointestinal complaints. Extraintestinal complications are common but usually follow the onset of the bowel complaints. We present an unusual case in which scrotal swelling was the first symptom in a patient discovered to have Crohn's disease. This is particularly unusual in that the patient presented as a case of urethral fistula.

INTRODUCTION

Crohn's disease (CD) may involve the perineum, but this complication most frequently follows gastrointestinal complaints. Genitourinary manifestations of CD are not common as presenting symptoms [1].

We present an unusual case of scrotal and penile involvement of CD with a complete MRI exploration. To our knowledge, no prior reported cases of this entity with MRI were reported previously.

CASE REPORT

A 48-year-old man presented with a 2-month history of scrotal swelling and pain. He has a history of CD involving the small intestine and colon since 1989, confirmed by colonoscopy and rectal biopsy. He was operated on 2 times for ileal obstruction. Mesalazine was administered thereafter. Under antibiotics, the swelling and pain decreased with the onset of purulent discharge.

A physical examination revealed 2 productive fistulas located in the scrotum and the perianal region. There was no evidence of induration or erythema. Compression adjacent to the orifice resulted in the expulsion of purulent material. The 2 testes were palpable without any clinical abnormality, and the digital rectal examination was unremarkable. Laboratory investigations, including hematologic and biochemical examinations, revealed no abnormalities, especially the white blood cell count, which was normal. The rectoscopy, performed to rule out an eventual subclinical rectitis, was normal. Scrotal ultrasonography found a normal aspect of the 2 testes without any deep collection.

MRI showed a complex perineal fistula with a sagittal trans-sphincteric tract involving the corpus cavernosum (Figure 1, Figure 2). There was no evidence of thrombosis within the corpora. There were 2 external openings, one in the scrotum and the other in the perineum, and at least 2 internal openings in the external anal sphincter. The low fistula into the anal canal had penetrated the root of the scrotum and the bulb of the urethra, extending into the penile shaft. There was no involvement of the lower urinary tract (bladder, prostate, and urethra). Urethroscopy didn't show any urethral narrowing

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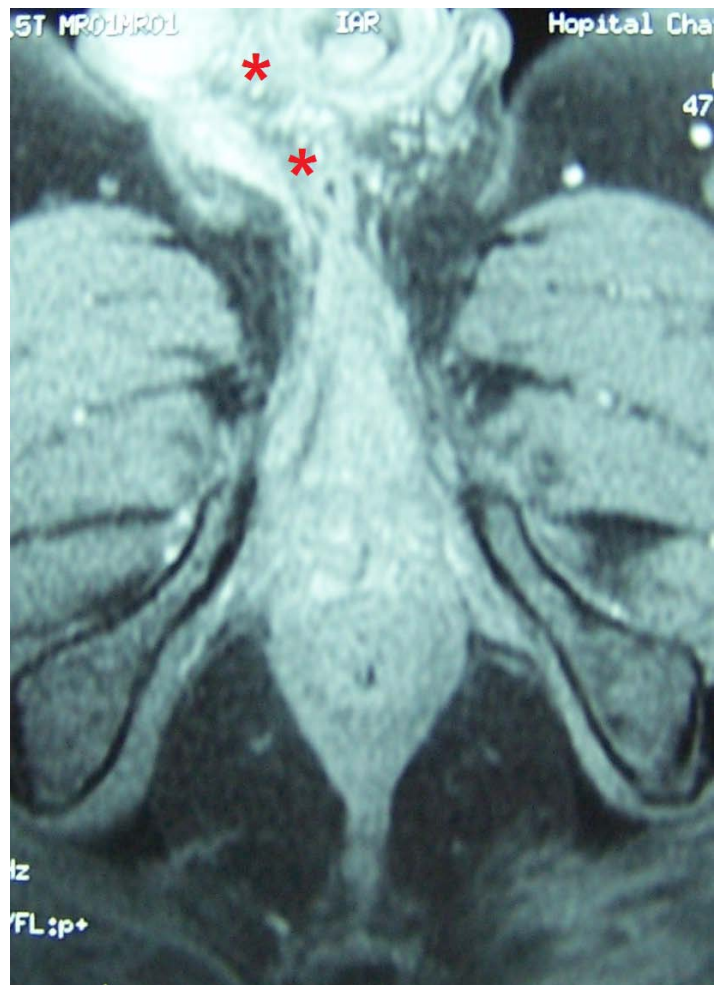
Figure 1. MRI of pelvis shows well-circumscribed abscess with central area of low attenuation and peripheral rim enhancement.

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Figure 2. MRI of the pelvis fitula within the left corpus cavernosum.

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or internal fistula opening. Thus, the diagnosis of complex fistulas with scrotal and penile involvement was made. Seton placement, associated with antibiotics as adjuvant treatment, was performed. Infliximab has been administered thereafter because of the complex aspect of fistulas. The patient's symptoms resolved and he was discharged.

Six months after, he remains in good health and has had no further relapses of his CD. The scrotal and perianal fistulae remain healed. He didn't report any gastrointestinal, voiding, or sexual complaints.

DISCUSSION

CD commonly presents in adults. Genitourinary manifestations of CD are rare and often secondary to gastrointestinal symptoms. Such complications occur in 5 to 20% of patients [1]. Two theories [2,3] were reported to explain genital involvement of CD in males:

1. Direct extension from the diseased bowel with transmural fistulization; it may affect all pelvic organs causing fistulae, abscesses, edema, and ulceration.

2. "Metastatic" deposit granulomas and abscesses involving the penis, scrotum, seminal vesicles, and prostate; they may form independently from the diseased intestine or in the absence of active bowel disease.

Metastatic CD was described for the first time by Parks et al. [4]. Clinically, it presents as ulcers, papules, nodules, plaques, or crusts located most frequently on the extremities, flexures, and genitalia. It is necessary to exclude other granulomatous diseases such as cellulitis, erysipelas, intertrigo, sexually transmitted diseases, hidradenitis suppurativa, lichenoid eruptions, erythema nodosum, and primary and secondary lymphedema.

Penile involvement in cutaneous CD is uncommon and manifests in the form of non-healing ulcers or erosions with or without oedema [5,6]. The ulceration can lead to considerable destruction and distortion of the surrounding soft tissues.

Pelvic MRI becomes the referenced technique in the evaluation of perianal CD. There is a high sensitivity and specificity of 100 and 86%, respectively, in the detection of a fistula's primary tracts, and a high sensitivity and specificity of 96 to 97%, respectively, in the detection of abscesses [7,8].

MRI is particularly useful for complex perianal fistulas to detect deep sepsis and their relationship with the sphincter and the genitalia [9]. MRI provides information about perianal disease activity and assesses inflammatory changes in fistula tracks in response to treatment with Infliximab [10,11]. To our knowledge, this is one of the first reported cases of MRI findings for a patient with CD involving the penis.

Endoscopic exploration of the lower urinary tract is indicated in such cases to rule out any other cause of fistula or an associated complication, such as urethral stricture.

The treatment of perineal and genital cutaneous CD is controversial, and a number of therapies have been advocated. But first of all a rectoscopy should be performed to establish the existence of macroscopic inflammatory activity in the rectum [12]. Antibiotics are useful, although they have only been assessed in uncontrolled, open-label studies (evidence level 3; recommendation grade D) [13].

In recent years there has been a significant change in treatment with the incorporation of new drugs such as immunosuppressants (azathioprine), 6-mercaptopurine, methotrexate and cyclosporine, and more recently infliximab, which are combined with local surgical treatments (abscess

drainage and seton placement), such as our case, in order to preserve rectal anal function [14].

CD fistulae seem to be an operative challenge for the surgeon, as they tend to be associated with significant scarring and insufficient surrounding tissues to allow for a tension-free closure. For this reason, interposition of the tissue for good vascular supply is necessary to facilitate healing [15]. Fistulotomy will rarely be possible and, before establishing any surgical option, rectosigmoid intraluminal inflammatory bowel activity must be controlled [12]. In all cases, long-term medical treatment and follow-up are mandatory.

CONCLUSION

Genitourinary manifestations of Crohn's disease are rare. MRI can help determine disease activity in genitourinary locations and might be helpful in selecting a subpopulation of patients who should be monitored more closely for the development of more extensive disease.

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