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Primary syphilitic proctitis: case report and literature review

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Abstract

Rectal ulcerations are an uncommon presentation of a primary syphilis infection. Anorectal syphilis is difficult to diagnose because of its often asymptomatic or atypical clinical presentation. It is important to consider sexually transmitted diseases (STD) in all patients presenting with rectal symptoms. A history of anal sexual intercourse should be made, especially in men having sex with men (MSM). Moreover, the possibility of a primary syphilis infection of the rectum should be considered. Endoscopic findings might be diverse, whereas a typical chancre can present as an anorectal ulcer associated with regional lymphadenopathy. It is important to consider other causes of anorectal ulcers, like other STD, inflammatory bowel disease (IBD) or even malignant causes. The diagnosis of anorectal syphilis is based on the combination of the clinical presentation, serology tests, endoscopic findings and biopsies. The cornerstone of the treatment is based on an intramuscularly administration of a long-acting preparation of penicillin (benzathine penicillin G). (Acta gastroenterol. belg., 2018,

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Introduction

Patients often present with symptoms of diarrhea mixed with blood and mucus. The differential diagnosis of inflammatory bowel disease (IBD), infectious colitis, ischemic colitis etc. is well known by physicians. However, sexually transmitted diseases (STD) should also be considered. We report a case of proctitis due to a primary syphilis infection. In the discussion, we propose a general diagnostic work-up for patients presenting with new onset mushy stools mixed with blood and mucus and on overview of what is known about syphilitic proctitis.

Case history

A 36-year-old man with a family history of Crohn's disease was referred to the outpatient clinic by his general practitioner to exclude chronic inflammatory bowel disease with anorectal involvement. The patient complained about mushy stools mixed with blood and mucus during the last four days. At the same time, he mainly suffered from a perianal discomfort, abdominal pain and a fever (38.9 degrees Celsius). He had no prior important medical history. His sister was known with severe Crohn's disease with perianal manifestations. Further systemic history didn't reveal any important

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issues. A physical examination showed a mild abdominal tenderness in the right lower quadrant of the abdomen. Anal inspection showed no signs of an abscess or fistula. Adenopathies could not be clinically discovered.

Laboratory results demonstrated a normal white blood cell count (8.69 x10*9/l) with a normal differentiation, but an elevated C-reactive protein level (40 mg/l [0.0 – 5.0]). The remainder of the blood analysis was satisfying. We performed a stool analysis that could not identify any pathogens. Analysis of fecal calprotectin turned out to be highly positive (814 microgram / g feces, whereas a normal range is below 50).

The patient was admitted the same day in the hospital for a semi-urgent diagnostic work-up.

Taking in account the family history of IBD a magnetic resonance imaging (MRI) of the lower abdomen and pelvis was performed for the purpose of excluding fistulous abscesses. The images showed an edematous rectal abdominal wall with inflammatory infiltration of the mesorectal adipose tissue and the presence of enlarged lymphatic nodules (Fig. 1).

An ileocolonoscopy was performed the day after. There was a normal appearing mucosa of the terminal ileum and colon, except for the inspection of the rectal mucosa, which showed multiple atypical mucosal ulcerations with adjacent edematous mucosa (Fig. 2). Multiple biopsies were taken in the ileum, the colon and rectum. The anatomopathological findings of the terminal ileum and colon were normal. Those of the rectum showed a mild chronic inflammatory infiltrate with an excess of plasma cells, suggestive of an infectious colitis. There was no evidence for underlying IBD.

Taking in account that the rectal ulcers had atypical characteristics, we asked for sexual behavior. Because the patient admitted having unprotected anal intercourse, we performed testing for STD. The analyses of Chlamydia trachomatis and Neisseria gonorrhoeae on a rectal

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Fig. 1. — Magnetic resonance imaging (MRI) of the lower abdomen and pelvis shows an edematous rectal abdominal wall with inflammatory infiltration of the mesorectal adipous tissue and the presence of enlarged lymphatic nodules in the mesorectal adipous tissue.

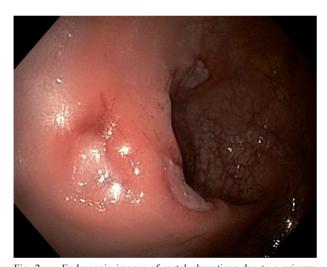


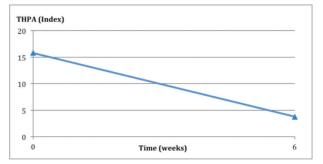
Fig. 2. — Endoscopic images of rectal ulcerations due to a primary

swab and on an urine sample (to exclude urethritis as a co-infection) were negative. We also performed a more general diagnostic work-up for other STD by screening for hepatitis B, hepatitis C and HIV by serologic tests. These results turned out to be negative. Laboratory analyses however demonstrated a positive Treponema pallidum hemagglutination assay (TPHA) (index 15.8, positive when > 1.1) and a positive Venereal Disease Research Laboratory test (VDRL).

We established the diagnosis of an early primary syphilis infection of the anorectum, also described as a syphilitic proctitis, and treated the patient and his sexual partner with one intramuscular injection of 2.4 million units of benzathine penicillin G.

An endoscopic reassessment was performed six weeks after initiation of the treatment. This showed a proper

Table 1. — Evolution of serological tests shows a decline of the TPHA titer after treatment, but TPHA still remains positive. TPHA, Treponema palidum hemagglutination assay



endoscopic healing with remaining small stellate scars. Control of serologic tests after treatment showed a negativation of the VDRL test while the TPHA test remained positive (Table 1).

DISCUSSION

The general diagnostic work-up in patients presenting with new onset mushy stools mixed with blood, mucus and fever starts with a broad history and clinical examination. It is important to ask for recent use or changes in medications (especially antibiotics, nonsteroidal inflammatory drugs etc.), dietary habits, recent travels abroad, associated red flags such as weight loss, familial history of IBD and past history of abdominal problems or surgical interventions. Perianal inspection should not be forgotten during a broad clinical examination as well as inspection and palpation for adenopathies. The first diagnostic intervention should consist of analysis of a blood sample with specific attention for inflammatory parameters. Analysis of stool cultures with a direct examination for parasites and ovae are necessary to exclude enteropathogens. A faecal calprotectin analysis is not recommended as a first line screening tool due to its high cost and it adds no extra value to the diagnostic work-up if an endoscopy is already performed. In our case, we performed a faecal calprotectin analysis taking into account the familial history of Crohn's disease. At the time of consultation, an anoscopy or rectoscopy should be performed and if necessary followed by an ileocolonoscopy after bowel preparation to exclude mucosal damage (as seen in infectious, inflammatory, ischemic colitis etc.). Multiple biopsies have to been taken for anatomopathological examination. In the unlikely case the colonoscopy turned out to be negative, further imaging by using CT or MRI to demonstrate an internally draining abscess, fistulae, strictures or to exclude mucosal lesions in the more proximal intestine is useful. Another indication to perform a CT (or MRI) scan of the abdomen as a logical first approach is in the presence of peritoneal signs in which case an endoscopy can be contraindicated. Because of the high cost and the lower availability of MRI, this is not a standard test to perform in the diagnostic work-up. If a STD is suspected, like in our

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case, additive tests are recommended, for example a rectal swab for Chlamydia trachomatis, Herpes simplex virus and Neisseria gonorrhoeae as well as serologic tests for Treponema pallidum. A broad screening for co-infections with hepatitis B virus (HBV), hepatitis C virus (HCV) and human immunodeficiency virus (HIV) is strongly advised.

In this case report, the diagnosis of a primary syphilis infection of the anorectum was made in a patient with unusual appearing rectal ulcerations after unprotected anal sexual intercourse. A literature search revealed only a few published similar cases of a primary syphilitic proctitis (1-3). The clinical presentation can be very atypical and therefore it is often a missed diagnosis. The presentation can be asymptomatic. Mild symptoms like anal pain, tenesmus, itching, anal discharge, rectal bleeding or urgency of defaecation can be present. Treponema pallidum may cause a proctitis. The endoscopic presentation of syphilitic proctitis can be broad. Ulcers can be multiple, irregular, two ulcers can be opposite to each other or they may be eccentrically located (4). Sometimes a typical papule appears at the site of inoculation after an average incubation period of two to three weeks instead of atypical ulcers. This papule ulcerates and there is an evolution to an ulcer with a raised and indurated margin that is the typical chancre of a primary syphilis. The ulcer has a non-exudative base. The chancre is normally associated with regional lymphadenopathies. After three to six weeks, the primary chancre will heal spontaneously and leaves an indurated

Proctitis due to a primary syphilis infection is often confused with IBD and rectal neoplasm (5). Therefore, it is important to ask for sexual history and to inform whether unprotected anorectal intercourse has taken place. This uncommon etiology should be suspected in young men having sex with men (MSM) (4). Other STD like herpes simplex virus, cytomegalovirus, Neisseria gonorrhoeae infections or lymphogranuloma venereum should also be suspected. Other non-infectious causes such as total rectal prolapse, invagination with solitary rectal ulcer syndrome, trauma caused by foreign bodies, Behçet's disease and a lymphoma should be considered in the broad differential diagnosis (6).

Diagnostic testing for syphilis infection consists of dark-field microscopy for treponemes or a nucleic acid amplification test for Treponema pallidum DNA from exudate out of an ulcerated lesion (6). Because of contamination from commensal spirochetes that are found in the normal rectal flora, the dark-field microscopy testing can be inaccurate. Serologic tests such as TPHA and VDRL support the diagnosis of syphilitic proctitis in the presence of symptoms or endoscopic signs of proctitis (6). The combination of serologic tests and biopsies of an anorectal lesion is the recommended way for making the diagnosis (6). The

endoscopist should perform multiple biopsies because the histological evaluation can often be nonspecific (6-7). A Warthin-Starry staining should be performed because this is known to be specific for syphilis and can identify spirochetes (6). Though, like in our case, this test can be difficult with a low sensitivity, and a negative result does not exclude a syphilis infection.

A treatment with an intramuscular administration of penicillin is the treatment of choice in the case of primary syphilis of the anorectum if there is no known penicillin allergy. Since Treponema pallidum divides slowly with an average of one doubling in vivo per day, low and continuous levels of penicillin are essential for the treatment and elimination of treponemes. Therefore, the use of the long-acting benzathine preparation, benzylpenicillum or penicillin G 2.4 million units in one intramuscular injection, is the initial treatment of choice. Treatment in patients with longstanding disease may require a dose repeat at weekly intervals for three weeks because of the possibility of a late latent syphilis infection instead of an early primary infection (2). Alternative ways of treatment in patients with a penicillin allergy are an oral treatment with doxycycline 100 mg twice a day for 14 days or tetracycline 500 mg four times daily during 14 days. Screening for other STD has to be performed and especially a co-infection with HIV has to be ruled out. Specific advice should be sought if the patient is HIV co-infected as treatment regimens may vary (4). It is important not to forget the source of infection and so the sexual partner or partners of the patient should also be proper treated with a single intramuscular injection of benzylpenicillum.

Follow-up should be performed by combining a rectal reexamination together with serologic testing at regular intervals, especially at six and twelve months after treatment (8).

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