

Unusual presentation of Crohn's disease in a boy with penile swelling

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Key Clinical Message

Crohn's disease is one of the three subtypes of inflammatory bowel disease, with regional, transmural, and granulomatous inflammation. The present study is a case report on an 11-year-old boy who was referred to the hospital due to penis swelling and pain since a year ago, the patient sometimes complained of pain when urinating, he was having a severe painful swelling in the penis shaft; however, his scrotum was completely normal. He was diagnosed with Crohn's disease, thus was placed on azathioprine therapy which eradicated his pain after 3 months, and thus, his skin lesion became much smaller. Crohn's disease, especially in children, should be considered in patient with unknown skin complications, and, if clinically suspected, biopsy examination should be performed. The diagnosis of Crohn's disease may be delayed in extradigestive cases. This disease occurs when associated with skin manifestations, which is uncommon. We reported a case with penile Crohn's disease, of which few cases have been reported in children to date.

KEYWORDS

Crohn's disease, inflammatory bowel disease, penile swelling

1 | INTRODUCTION

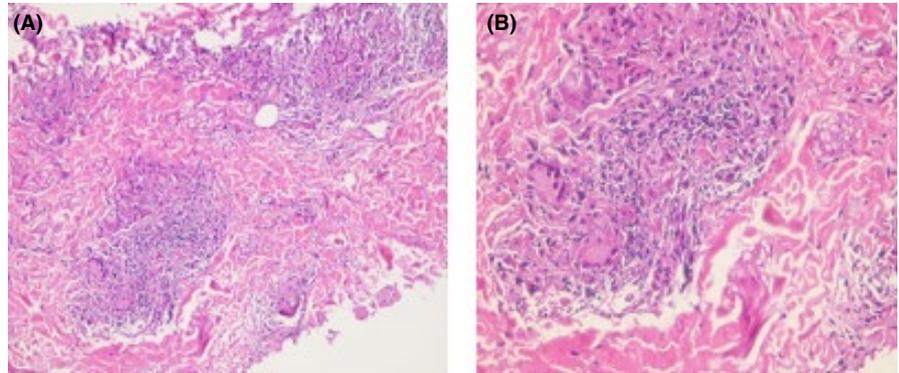
Crohn's disease (CD) and ulcerative colitis (UC) are the two main types of inflammatory bowel disease. CD also referred to inflammatory bowel diseases (IBDs), is gastrointestinal tract (GIT) immune-mediated, of unrecognized but possibly multifactorial etiology. Patients suffering from IBD (CD and UC) can as well be inflicted with manifestation outside the GIT; these extraintestinal manifestations (EIMs) for about 50 years prior have been described in the literature.¹ The most commonly affected parts of the body are the eyes, the joints,² and the skin, but numerous organs have been noted as potential locations and these include but not limited to the central nervous system, kidney, blood, skeleton, lungs, and the hepatobiliary system.³

Although CD is still being considered an incurable disease, various therapies have been attempted for the various clinical manifestations and complications of this disease. Recently, many treatment policies based on the results of clinical studies have been proposed.⁴ Here is a case with penile CD, few cases of which have been reported in children so far.

2 | CLINICAL REPORT

An 11-year-old boy was referred due to penis swelling and pain from 1-year prior, and sometimes he complained of pain when urinating. There was no history of trauma and sexual contact, abdominal pain, diarrhea, constipation, fever,

FIGURE 1 (A and B) Hematoxylin and eosin staining of punch biopsy of penile swelling showed multiple foci of lymphohistiocytic infiltration with granulomatous inflammation with no central necrosis consistent with cutaneous manifestations of crohn's disease (magnification $\times 200$)



arthritis, and skin problems in the patient. His height and weight were above the 50th percentile. There was no evidence of redness and inflammation symptoms in the whole skin examination, but there was a severe painful swelling in the penis shaft. The scrotum was completely normal (parents did not consent to take pictures at this stage of the disease). The abdomen, pelvis, heart, and lung examinations were completely normal. The tests results were as follow:

ESR: 28, CRP: 1, Hb: 12.5, plt: 228 000, WBC: 8500, stool examination; WBC: 3-4, RBC: 1-2, calprotectin: 153, P-ANCA: negative, ASCA: negative.

The patient had a 9-year-old brother who was diagnosed with CD and treated with azathioprine from 1-year prior (with a history of abdominal pain, weight loss, and bloody diarrhea). After the rejection of infectious and cutaneous tuberculosis (TB) problems and rejection of urological problems, and with suspicion of CD, a punch biopsy on the penile swelling was performed and numerous granulomas (using H&E staining with negative Zilnelson culture) were observed in the pathologic sample (Figure 1A,B) and penile CD was finally raised for him. The digestive system of the patient was examined with esophageal gastroduodenoscopy and colonoscopy, and multiple biopsies were taken, the appearance and pathology of which were completely normal. Considering that there is no single specific treatment for CD today, on the one hand, and, on the other hand, drugs such as infliximab are considered as the last line therapy, which sometimes causes serious and irreparable complications in children; also considering the fact that a satisfactory therapeutic response to azathioprine was observed in the patient's brother, so azathioprine therapy was given to the patient, and thus, relieving his pain and his skin lesion became much smaller (parents at this stage were content to take pictures) (Figure 2).

3 | DISCUSSION

Crohn's disease is a chronic inflammatory and granulomatous disorder of the gastrointestinal tract that occurs at any age, but most especially at the second decade of patient's life.



FIGURE 2 After 3 mo of administration of azathioprine, penile swelling decreased

Children with CD may have a wide range of symptoms. It is very important to get familiar with the main and uncommon symptoms of the disease.⁵ Some children may develop the disease with nontypical symptoms which may delay the diagnosis and commencement of treatment.⁶ About 25% of the patients may have a nondigestive symptoms manifestation, including short stature and growth retardation, joint involvement (arthritis and arthralgia), cutaneous involvement (pyoderma gangrenosum, erythema multiforme, etc), liver, and bile dysfunction, etc.^{5,7}

Uncommon cases of CD cutaneous involvement are seen in children.^{6,8,9} Cutaneous CD affecting the perineum, vulva, and perianal skin is an uncommon entity, which may precede or accompany gastrointestinal CD. If vulva is involved and left untreated, an extensive surgery may be required such as vulvectomy to gain control of the disease. Both cutaneous and gastrointestinal CDs respond to biologics blocking TNF α . Furthermore, ustekinumab, IL-12 and IL-23 cytokines target, is potent in patients with gastrointestinal CD who fail TNF blockade. However, whether ustekinumab is potent for cutaneous CD is yet debatable.¹⁰

Until now, there has been no single treatment for this disease, and there have been various treatments for it in different reports; also given that the patient's brother (who had intestinal CD) had a good response to azathioprine, azathioprine was recommended to the patient as well. The pain and swelling of the penis was significantly reduced within 3 months of the onset of treatment. Until this time, the patient is monitored for other digestive and extradigestive symptoms of CD. Since the cutaneous involvement is an uncommon manifestations of CD, especially in children; therefore, this disease must be considered in patient with unknown skin complications, and, if clinically suspected, biopsy examination must be performed.

CONFLICT OF INTEREST

All authors report no conflicts.

AUTHOR CONTRIBUTION

FM: Conception and design, acquisition of data or analysis and interpretation of data. BE: Drafting the article or revising it critically for important intellectual content. SA: Final approval of the version published, agreement to be accountable for the article and to ensure that all questions regarding the accuracy or integrity of the article are investigated and resolved.

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