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Erythema Nodosum, the First Clinical Manifestation of Crohn's Disease in a-14 Year-Old Boy: A Case Report

Sheila Kianifar (MD)[1], Abdolreza Malek (MD)[2*], Mohammad Ali Kiani (MD)[2], Hamid Reza Goldouzi (MD)[1]

[1]*Resident of Pediatrics, Department of pediatrics, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.*
[2]*Department of Pediatrics, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.*

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ABSTRACT

Erythema nodosum (EN) is a condition in which the skin suffers from oval-shaped and tender erythematous patches, mostly on the anterior region of the leg and sometimes other parts of the body, including the arms and affects subcutaneous adipose tissue. These patches are self-limiting and usually heal on their own within one to two weeks. This is a case report of a patient who is a 14-year-old boy complained of oval-shaped tender erythematous lesions on the legs of both legs and referred and was hospitalized 5 days ago. The patient continued to complain of pain in both thighs when standing and sitting. The patient developed a high fever upon admission. There was no history of the disease while we were taking his history. All laboratory tests were performed and rheumatic diseases and possible malignancies were evaluated and rejected. The patient was treated with the anti- naproxen, an inflammatory drug, and then the patient's pain and fever decreased. However, the patient suffered from multiple ulcerative lesions in the mouth on the final day of hospitalization. Infectious counseling was performed and he was discharged with a diagnosis of EN in the context of oral herpes. Two days after discharge, the patient suffered from hematochezia. Consequently, he was hospitalized and underwent colonoscopy, which showed multiple mucosal lesions in the esophagus and duodenum, and a sample was sent for pathology. The test result showed Crohn's disease, and the patient was treated immediately.

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Introduction

Erythema nodosum is a condition in which the skin suffers from oval-shaped and tender erythematous patches, mostly on the anterior region of the leg and sometimes other parts of the body, including the arms, which is also seen as oval in the involvement of subcutaneous adipose tissue(1, 2).

These patches are self-limiting and usually heal on their own within one to two weeks. These erythematous lesions vary in size from 1 to 6 cm and are initially red in color and gradually turn brown or purple, with a circular or oval shape. The etiology of these lesions is unknown in 30% to 50% of cases, and is associated with an infectious or rheumatic or gastrointestinal disease, including inflammatory A disease in other cases. Infectious diseases associated with EN include viral diseases such as hepatitis B and epstein-barr virus , bacterial diseases including tuberculosis, Yersinia, group A strep, Cat-scratch disease, Whipple's disease, brucellosis, tularemia, and mycoplasma. Other systemic diseases include sarcoidosis, IBD, and the use of medications including oral contraceptives, behçet's disease, severe acne, Hodgkin's disease, and pregnancy. The etiology is

***Corresponding author:** Abdolreza Malek, Department of Pediatrics, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.*
***E-mail:** malekar@mums.ac.ir*
***Tel:** 09151037242*

unknown in 30% to 50% of cases (3). EN is probably a hypersensitivity reaction Ato the myriad antigens present in various diseases. Circulating immune complexes have been identified in IBD in these cases but not in idiopathic cases (4). EN has been reported in 6% to 15% of patients with Crohn's disease and 1% to 4% of patients with ulcerative colitis (3), but these cases have been very few (4).

Treatment of the underlying disease should be accompanied by symptomatic treatment. Pharmacological treatments include salicylates, nonsteroidal anti-inflammatory drugs, colchicine, and intralesional steroid injections for idiopathic and limited lesions. For recurrences, a complete laboratory examination is needed to rule out the causes of malignancy and other systemic causes, including: complete blood count, blood sedimentation rate, pharyngeal culture, tuberculosis test, CRP, and chest x-ray. Statistics show that patients with IBD have one or more gastrointestinal symptoms in 6% to 40% of cases (3, 4) and patients with IBD have skin symptoms in 15% of cases. EN is the most common form of skin involvement in IBD, which includes 3% to 10% of patients with ulcerative colitis and 4% to 15% of patients crohn's disease (5,6).

These subcutaneous lesions are often prominent, red or purple in color and reach about 1 to 5 cm. These nodules are often seen on the extensor surfaces of the limbs, especially on the anterior surface of tibia. Regional panniculitis is visible in biopsy specimen of these lesions. However, the diagnosis is often made clinically and the biopsy is performed only in atypical cases, such as cases where the lesion is not on the leg or lesions persist more than 6 to 8 weeks or become injured. Skin manifestations are often associated with gastrointestinal manifestations, and underlying disease treatment often leads to the disappearance of lesions. In rare cases, skin lesions occur before or during gastrointestinal manifestations. In these cases, EN treatment is performed independent of treatment of the underlying disease and prednisolone courses may be required (7).

Case presentation

The patient is a 14-year-old boy referred and admitted with complaint of oval-shaped tender erythematous lesions on the tibia of both legs from 5 days ago.(Fig-1) The patient still complained of pain in both thighs when standing and sitting. The patient developed a high fever upon admission. There was no history of the disease. All laboratory tests were performed and rheumatic diseases and possible malignancies were evaluated and rejected. Also, various skin, infectious disease, and allergy consultations were performed for the patient.

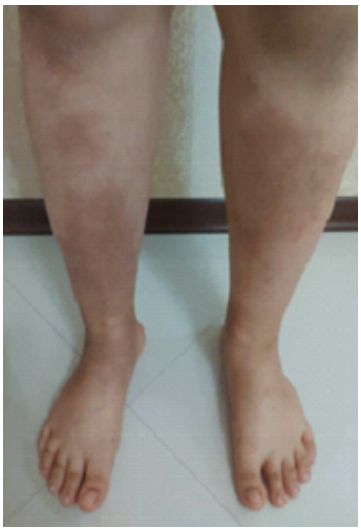

Figure 1: Erythema nodosum.

Other examinations were normal and there was no sign of arthritis or cellulite on examination of the organs. There was only a slight tenderness in the thighs. The result of bone marrow aspiration procedure was normal, and skin biopsy showed the EN pathology. The patient underwent naproxen treatment, an anti-inflammatory drug. Afterward, the patient's pain and fever decreased, but the patient suffered from several ulcerative lesions in the mouth on the final day of hospitalization. Infectious disease consultation was performed and he was discharged with a diagnosis of EN in the context of oral herpes. Two days after discharge, the patient suffered from hematochezia. Consequently, he was hospitalized and underwent colonoscopy, which showed multiple mucosal lesions in the esophagus and duodenum, and a sample was sent for pathology. The test result showed Crohn's disease, and the patient was treated immediately.

The result of laboratory tests is as follows:
WBC: 15400 (P: 78 ,L:22) , Hb: 13.4 mg/dl , Plt: 616/mm3 , ESR: 95mm, CRP: 74 mg/dl,
AST: 24 mg/dl, ALT: 17 mg/dl, Urea: 8 mg/dl, Cr: 0.9 mg/dl, LDH: 371 mg/dl,
Uric acid 4.4 mg/dl, NA: 128 → 140 meq/L, k: 2.8→ 3 meq/L,
COLD AUG: Neg, IGM Anti VCA: Neg, MONO SPOT: Neg ,
FANA: Neg, RF: Neg, CANCA: Neg, PANCA: Neg,
Blood culture: Neg U/A: Normal U/C: Neg,
WRIGHT , 2ME: Neg, Serum ACE: Neg, PPD Test: Neg, , ASO: 100 Todd.

Most cases of Crohn's disease occur with gastrointestinal manifestations and there are a few cases that have been manifested in the form of

skin problems. This patient was selected to introduce rare and unusual manifestations of the disease and how to diagnose and treat the patient based on symptoms.

Discussion

Erythema nodosum is circular or oval tender, red or purple lesions that appear on the anterior surface of tibia and sometimes the arms. The size of these lesions is 1 to 6 cm and affects the subcutaneous adipose tissue (7).These lesions are the most common form of skin involvement in IBD and are associated with gastrointestinal manifestations in a way that the treatment of the underlying disease is also effective in improving the symptoms of these lesions. The disease is unknown in 30% to 50% of cases and is often self-limiting and resolves within 1 to 2 weeks. Numerous causes have been suggested for this problem, including infectious, rheumatic, and malignant diseases (3).

EN is a disease of increased sensitivity in response to various antigens and is observed in 6% to 15% of patients with Crohn's disease and 1% to 9% of those with ulcerative colitis (4). A study was performed on 3266 patients with IBD, of which 354 had Crohn's disease and 136 had ulcerative colitis with skin manifestations. Crohn's disease was most commonly seen in young women, and a positive family history of the disease also played an important role in this regard. In this study, patients with skin involvement needed more antibiotic, steroid, and immunosuppressive therapy (7).

In a study conducted on inpatients with EN at Soroka University between the years 2004 and 2014, the most common symptoms included arthritis or arthralgia (8). A study investigated skin involvement in vulvar and perineal areas in children. This study evaluates 20 studies and found only 22 patients with vulvar Crohn's disease. The above study also showed that the disease is uncommon and difficult to diagnose due to nonspecific symptoms (9). The most common manifestations of chronic IBD, except gastrointestinal involvement, include cutaneous, musculoskeletal and hepatobiliary manifestations (10).

A cohort study comparing incidence of pediatric IBD and (<16) and adult IBD showed that pediatric IBD led to higher risk for the need for immune and biological regulatory drugs to treat and control the disease (11). In one case, the patient was an 11-year-old boy suffering from Crohn's disease. He was referred with a complaint of swelling and pain in the penis from a year ago, which has been reported as a very rare manifestation of Crohn's disease (5). In cases of spondyloarthritis, which occurs in the context of IBD, clinical or subclinical intestinal inflammation has also been observed (12). A 20-year study revealed that more than one in six patients with IBD showed symptoms of arthritis (13).

In the patient introduced, the first IBD manifestation was EN in a 14-year-old boy, which occurred before the onset of gastrointestinal symptoms. This is a rare case, and the patient was diagnosed by colonoscopy after hematochezia. Reporting these cases of patients with rare manifestations such as the one seen in the patient introduced in the present study can pave the way for the rapid diagnosis of similar cases and early treatment of patients in the future.

Conclusion

According to the results in children with EN, many differential diagnoses must be considered at the same time. Otherwise, serious illnesses that present with this symptom can pose many risks due to delayed diagnosis.

Conflict of Interest

The authors declare no conflict of interest.

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