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Case Report

Chronic Recurrent Multifocal Osteomyelitis Associated With Inflammatory Bowel Disease Successfully Treated With Infliximab

Shinhyeung Kwak,[1] **Dongsub Kim**,[1,2] **Joon-sik Choi**,[1,3] **Yoonsun Yoon**,[1,4] **Eun Sil Kim**,[1] **Mi Jin Kim**,[1] **So-Young Yoo**,[5] **Jong Sup Shim**,[6] **Yon Ho Choe**,[1] **Yae-Jean Kim**[1]

[1]Department of Pediatrics, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, the Republic of Korea
[2]Department of Pediatrics, Kyungpook National University School of Medicine, Daegu, the Republic of Korea
[3]Department of Pediatrics, Yongin Severance Hospital, Yonsei University College of Medicine, Yongin, the Republic of Korea
[4]Department of Pediatrics, College of Medicine, Korea University, Seoul, the Republic of Korea
[5]Department of Radiology, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, the Republic of Korea
[6]Department of Orthopedic Surgery, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, the Republic of Korea

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Correspondence to
Yae-Jean Kim
Division of Pediatric Infectious Diseases and Immunodeficiency, Department of Pediatrics, Samsung Medical Center, Sungkyunkwan University School of Medicine, 81 Irwon-ro, Gangnam-gu, Seoul 06351, the Republic of Korea.
Email: yaejeankim@skku.edu

ORCID iDs
Shinhyeung Kwak
https://orcid.org/0000-0002-6166-2361
Dongsub Kim
https://orcid.org/0000-0002-9836-6769
Joon-sik Choi
https://orcid.org/0000-0002-5587-2960
Yoonsun Yoon
https://orcid.org/0000-0003-0187-3922
Eun Sil Kim
https://orcid.org/0000-0003-2012-9867

ABSTRACT

Chronic recurrent multifocal osteomyelitis (CRMO) is an inflammatory bone disorder presenting with sterile osteomyelitis, most often presenting in childhood. Although the etiology is understood incompletely, its association with other auto-inflammatory diseases including inflammatory bowel disease (IBD); psoriasis; Wegener's disease; arthritis; and synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome suggests that dysregulated innate immunity may play an important role in the pathogenesis. We report a case of a 13-year-old boy with CRMO associated with Crohn's disease (CD) successfully treated with infliximab after failure of non-steroidal anti-inflammatory drug (NSAID) treatment. He initially was diagnosed with CRMO based on symmetric and aseptic bone lesions with no fever, lack of response to antibiotic treatment, vertebral involvement, and normal blood cell counts. Despite five months of NSAID treatment, his musculoskeletal symptoms were aggravated, and he developed gastrointestinal symptoms. Finally, he was diagnosed with CRMO associated with CD. Due to the severity of symptoms, infliximab was initiated and produced symptom improvement. This case supports infliximab as another choice for treatment of bowel symptoms in addition to the bone and joint symptoms of CRMO when other first-line treatments are ineffective.

Keywords: Chronic recurrent multifocal osteomyelitis; Inflammatory bowel disease; Crohn's disease; Infliximab

INTRODUCTION

Chronic recurrent multifocal osteomyelitis (CRMO; also known as chronic nonbacterial osteomyelitis, CNO) is a rare inflammatory disorder presenting with bone pain from sterile osteomyelitis. It was described first in 1972 as subacute and chronic symmetrical

Mi Jin Kim
https://orcid.org/0000-0002-4505-4083
So-Young Yoo
https://orcid.org/0000-0002-8203-3441
Jong Sup Shim
https://orcid.org/0000-0002-5472-419X
Yon Ho Choe
https://orcid.org/0000-0003-1525-7688
Yae-Jean Kim
https://orcid.org/0000-0002-8367-3424

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osteomyelitis.[1] There are currently about 400 cases of CRMO described in the literature, predominantly as case series. However, accurate prevalence of CRMO is difficult to assess due to poor awareness of this rare disease and the possibility of its underdiagnosis. Rarely, CRMO can be accompanied by other autoimmune conditions such as inflammatory bowel disease (IBD) or psoriasis vulgaris, which complicate management. Here, we report a case of CRMO associated with Crohn's disease (CD) that was treated successfully with infliximab.

CASE

A 13-year-old Caucasian boy from Central Asia presented with a history of recurrent joint pain and multifocal painful soft tissue swelling during the preceding 18 months. He had pain in the right knee, both feet, and left great toe. Acute tenosynovitis and abscesses were suspected on both feet, for which he underwent eight operations of incision-and-drainage, followed by multiple courses of antibiotic treatment in other hospitals. His symptoms improved for a while, but pain recurred, for which he visited Samsung Medical Center (SMC) for further evaluation.

At the first visit to the SMC pediatric infectious disease clinic, he could no longer ambulate due to mild contracture of both ankles after repetitive surgical debridement. He did not present with fever, skin rash, abdominal pain, or gastrointestinal symptoms. His body gauge was the lower normal limit for his age; 36.3 kg, 1–3 percentile of his body weight, 151.2 cm, 5–10 percentile of the height, and the body mass index (BMI) was 15.88 kg/m^2, 3–5 percentile. Initial laboratory findings showed elevated erythrocyte sedimentation rate (ESR) of 76 mm/h, whereas white blood cell (WBC) count, C-reactive protein (CRP), and immunoglobulin levels were normal (**Fig. 1**). Cultures from blood and joint fluid were all negative. Magnetic resonance imaging (MRI) of the lower extremities showed symmetric inflammatory arthritis with marginal sclerosis on both ankles and feet with multifocal bone lesions (**Fig. 2A and B**). Increased radioactive uptake was observed on whole-body bone scan, suggesting vertebral involvement (**Fig. 2C**). Based on his history, clinical features, laboratory results, and radiologic findings, he was diagnosed with CRMO. He started naproxen treatment, and his condition stabilized enough to return to school.

After 5 months of naproxen (500 mg twice daily), his condition became aggravated with increasing pain and swelling in both ankles and psoriatic skin lesions on the scalp and both malleoli (**Fig. 3A and B**). He also developed abdominal pain with chocolate-colored diarrhea and revisited SMC for additional evaluation. Investigation showed anemia of chronic disease and iron deficiency; elevated ESR, CRP, and fecal calprotectin; and decreased albumin level (**Fig. 1**). A follow-up MRI showed extensive bone marrow signal change with enhancement in both femur and tibia (**Fig. 3C**) and a large subcutaneous abscess-like fluid collection (**Fig. 3D**), for which he received emergent incision-and-drainage. For the gastrointestinal symptoms, he underwent upper and lower gastrointestinal endoscopy and magnetic resonance enterography (MRE). On colonoscopy, cobblestone appearance with deep ulceration was observed from the cecum to the proximal descending colon (**Fig. 4A and B**). MRE revealed severe wall thickening with diffusion restriction from the ileocecal valve and proximal pelvic ileum, suggesting a chronic active stage of IBD.

As a result, the patient was diagnosed with CRMO with additional features of CD and psoriasis. Patients who have persistent bone pain and hyperintense signal within bone

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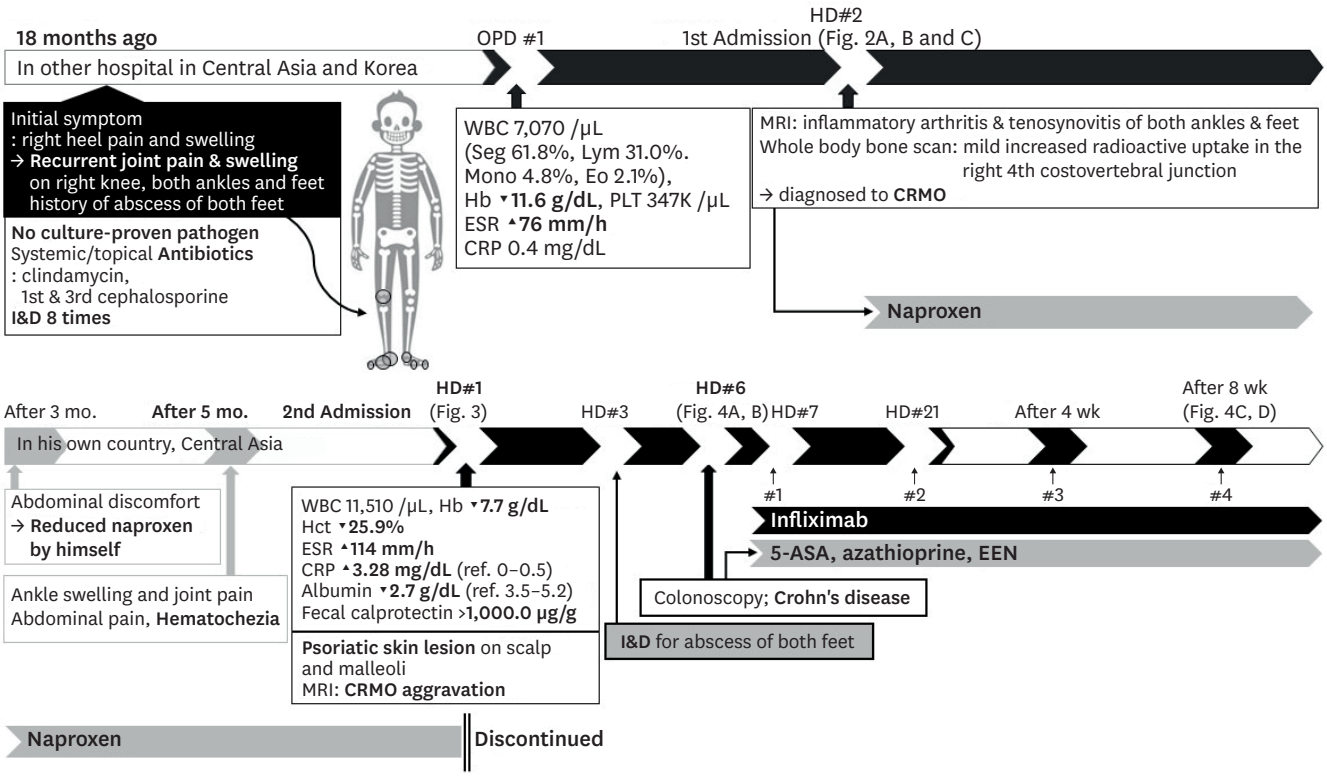

Fig. 1. Flow chart of the clinical course.
Abbreviations: OPD, outpatient clinic department; HD, hospital day; I&D, incision and drainage; WBC, white blood cell; Seg, segmented neutrophil; Lym, lymphocyte; Mono, monocyte; Eo, eosinophil; Hb, hemoglobin; Hct, hematocrit; PLT, platelet; ESR, erythrocyte sedimentation rate; CRP, C-reactive protein; MRI, magnetic resonance imaging; CRMO, Chronic recurrent multifocal osteomyelitis; 5-ASA, 5-acetylsalicylic acid; EEN, exclusive enteral nutrition.

marrow on imaging studies after three months of NSAID treatment are considered NSAID treatment failures. They need treatment with a second-line agent. Also, the aggravated CRMO condition with newly observed gastrointestinal symptoms despite naproxen treatment over three months was considered a failure of treatment for this patient. Therefore, he received infliximab as a second-line treatment, which is regarded to be effective for both CRMO and CD. As there was no definite guideline for the dose of infliximab in CRMO, the patient was administered infliximab according to dosage guidelines for CD. He received intravenous infliximab 5 mg/kg/dose at weeks 0, 2, and 6 for induction and 5 mg/kg/dose was planned every 8 weeks as maintenance. In addition, we added 5-aminosalicylic acid, azathioprine, and exclusive enteral nutrition (EEN), which are widely applied as a treatment protocol for CD. After induction therapy of infliximab, all musculoskeletal, gastrointestinal, and skin conditions improved dramatically. We used the pediatric Crohn's disease activity index (PCDAI), a multi-item instrument consisting of history, physical examination, growth parameters, and laboratory tests, for stratifying disease activity of Crohn's disease. PCDAI score ranges from 0 to 100 and the higher score means the more severe disease activity. PDCAI scores of 30 or higher indicate moderate to severe disease, and a decrease of 12.5 points or greater reflects the clinically significant response to therapy. His PCDAI was 62.5 at admission and improved to 22.5 after the third infliximab infusion. In addition, his colonoscopic findings also improved (**Fig. 4C and D**). He completed a 4th infliximab administration with significant improvement but was lost to follow-up after returning to his home country. This study was approved by the Institutional Review Board of Samsung Seoul Hospital (IRB No. 2021-08-122).

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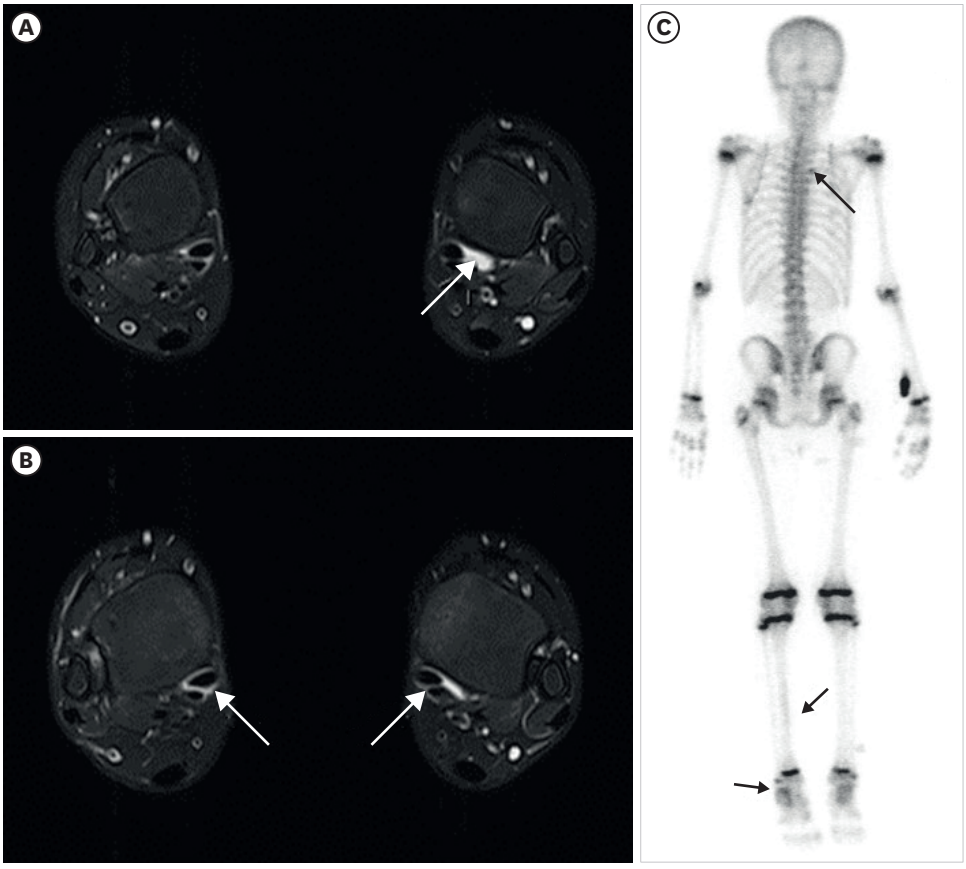

Fig. 2. Initial MRI and whole-body bone scan images. (A, B) Initial MRI of the lower extremities suggesting probable inflammatory arthritis associated with tenosynovitis (arrow) involving both ankles and feet with secondary atrophic change. (C) Whole-body MRI bone scan showing mild increased radioactive uptake in the right 4th costovertebral junction (arrow) with asymmetric uptake in left tarsal bone and tibia (white arrows). Abbreviations: MRI, magnetic resonance imaging.

DISCUSSION

We present the case of a 13-year-old boy diagnosed with CRMO with IBD that was treated successfully with infliximab. CRMO appears to have the highest prevalence in Western nations based on the current literature, and no global epidemiologic study has been reported.[2] It is primarily a pediatric disorder that can persist into adulthood or have an adult-onset presentation and is more frequent in girls than boys (female to male ratio of 2:1).[3] Typically presenting as insidious bone pain, CRMO most commonly affects the metaphyseal regions of long bones with sterile blood cultures and laboratory results demonstrating elevated ESR and CRP. Bone lesions can be visualized on MRI, which is an essential diagnostic tool. Empiric antibiotic therapy does not improve symptoms. CRMO is misdiagnosed commonly as the condition is obscure to many doctors, and when diagnosed, CRMO is concluded only after exclusion of other diseases. The most common differential diagnoses are infective osteomyelitis, malignancy such as Ewing's sarcoma, Langerhans' cell histiocytosis, juvenile idiopathic arthritis, non-specific musculoskeletal disorder, and viral infections.[4]

Additional coexisting conditions, including IBD, psoriasis vulgaris, and palmar plantar pustulosis, can occur with CRMO. The association of CRMO and IBD was mentioned for

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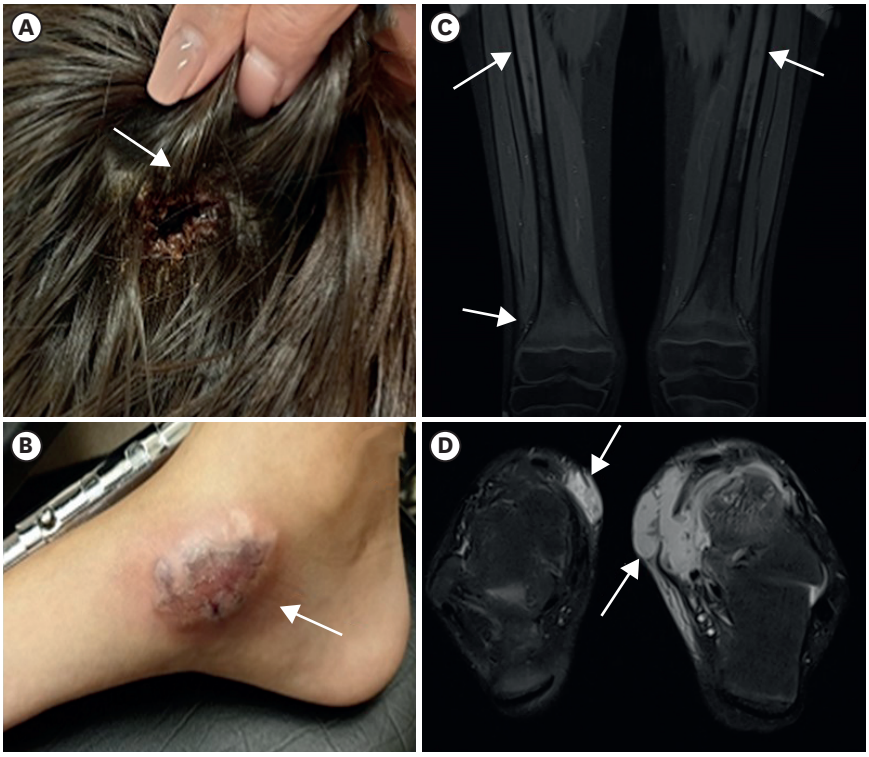

Fig. 3. Medical photos and MRI findings after five months of naproxen treatment. (A, B) New psoriatic skin lesion on the scalp and painful swelling of the right medial malleolus. (C) MRI suggesting newly noted extensive bone marrow signal change with enhancement in both femur and tibia, (D) inflammatory arthritis involving both ankles and feet associated with tenosynovitis with signal change in marrow of left talus. The arrows indicate large loculated and multiseptated subcutaneous abscess-like fluid collection invading muscle.
Abbreviations: MRI, magnetic resonance imaging.

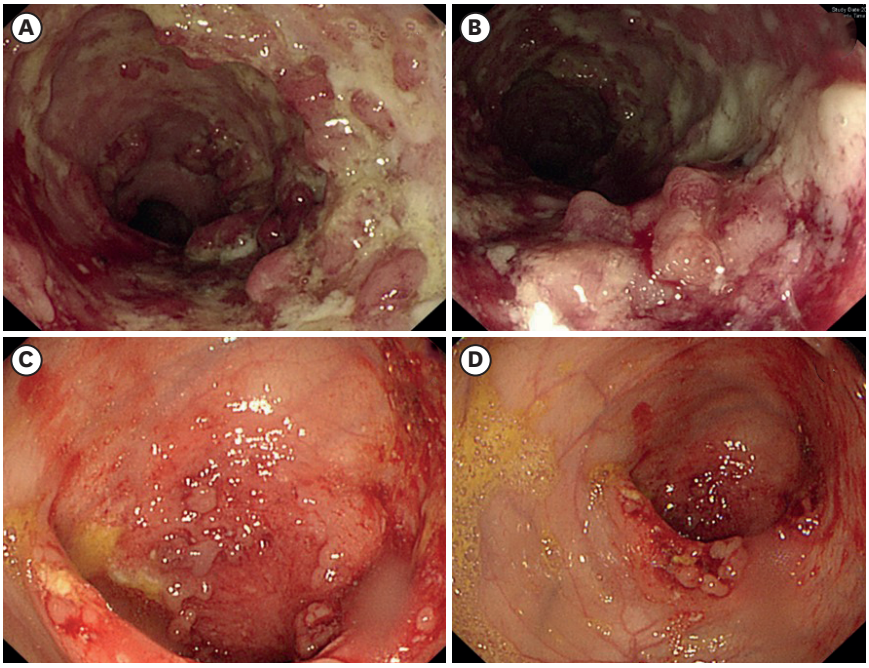

Fig. 4. Colonoscopy before and after infliximab therapy. (A, B) Initial colonoscopy revealing Crohn's disease; prominent cobblestone appearance with longitudinal deep ulceration observed from the cecum to the proximal descending colon. (C, D) Follow-up colonoscopy 8 weeks after end of infliximab induction therapy showing improved cobble stone appearance with ulceration remaining from the ascending to proximal transverse colon.

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the first time by Kahn, in 1990, who reported in a letter that, of 30-treated CRMO cases, five had IBD, including four cases of CD and one case of ulcerative colitis.[5] The first published association of CRMO and IBD was reported by Bognar et al. in 1998.[1] Since then, several cases of CRMO in association with IBD have been reported from various races and regions. **Table 1** summarizes the cases of CRMO associated with IBD that have been published in the literature. Although the affected patients were primarily children, several adult patients were reported.

CRMO recently has been classified as an autoinflammatory disorder rather than an autoimmune disease. These inflammatory conditions are characterized by episodes of systemic inflammation, including serological signs of inflammation occurring in the absence of autoantibodies, pathogens, and antigen-specific T cells. Components of the

Table 1. Summary of reported CRMO cases associated with IBD

Reference	No.	Sex/Age	Time of IBD onset (from CRMO onset)	IBD	Treatment for CRMO	Treatment for IBD
Current case	1	M/13	After 2 years	CD	NSAID → **IFX**	**IFX**, 5-ASA, azathioprine, EEN
Bognar et al. (1998)[1]	1	M/43	Simultaneous	CD	NSAID	Steroid, 5-ASA
Omidi and Siegfried (1998)[6]	1	F/12	NA	UC	Steroid	None
Bousvaros et al. (1999)[7]	6	F/8	After 3 months	CD	NSAID	Steroid, 5-ASA
		M/8	After 5 years	CD	None	Steroid, 5-ASA
		F/10	After 5 years	CD	Steroid	5-ASA
		M/10	After 3 years	CD	NSAID	Steroid, 5-ASA
		F/10	After 1.5 years	CD	None	Steroid, 5-ASA, MTX
		F/13	After 6 months	CD	None	Steroid, azathioprine
Bazrafshan and Zanjani (2000)[8]	1	F/12	Before 1 year	UC	NSAID	5-ASA, steroid → total colectomy
Huber et al. (2002)[9]	3		NA	NA	NA	NA
Schilling and Marker-Hermann (2003)[10]	5	F/9	After 4 years	CD	NSAID, steroid, calcitonin	5-ASA, steroid, azathioprine, MTX
		F/20	After 6 years	CD	NSAID	5-ASA, steroid
		M/33	NA	CD	NSAID	5-ASA
		F/39	After 7 years	UC	NSAID	5-ASA
		F/58	After 4 years	CD	NSAID	5-ASA
Carpenter et al. (2004)[11]	1	F/9	After 21 months	CD	NSAID	Steroids, metronidazole, 6-MP → azathioprine → **IFX**
Girschick et al. (2007)[12]	1	F/9	After 15 months	CD	NSAID, 5-ASA, steroid	Steroids, 5-ASA, azathioprine
Jansson et al. (2007)[13]	5		NA	2 CD	NA	NA
				3 UC		NA
Morbach et al. (2010)[14]	4	F/10	After 5 months	CD	NA	NA
		M/12	Simultaneous	CD	NA	NA
		F/15	Simultaneous	UC	NA	NA
		M/15	After 3 years	CD	NA	NA
Kim et al. (2012)[15]	1	M/41	Before 7 months	UC	NSAID, steroid, pamidronate, MTX	5-ASA, total colectomy
von Kalle et al. (2013)[16]	1		NA	CD	NA	NA
Ommen et al. (2015)[17]	1	M/10	After 1 month	CD	NSAID	Steroid, EEN → **IFX**, azathioprine
Audu et al. (2015)[18]	3	M/9	After 1 month	UC	None	Steroid, 5-ASA
		F/10	After 2 years	CD	Steroid	None
		M/2	After 3 years	CD	IVIG → steroid	5-ASA → EEN, azathioprine
Ahmed and Alsaleem (2019)[19]	1	F/11	After 6 months	UC	Steroid	5-ASA, azathioprine
Fujisaki et al. (2020)[20]	1	M/13	Simultaneous	CD	**IFX**	Steroid, 5-ASA, partial EN
Ng et al. (2021)[21]	1	F/12	After 2 years	UC	Bisphosphonate	Steroid, azathioprine
Total	37	M: 11	Simultaneous: 4	CD: 23	**Treatment with IFX: 4 cases**	
		F: 17	IBD precedent: 2	UC: 11		
		NA: 9	CRMO precedent: 20	NA: 3		
			NA: 11			

Reference, first author and reference number. Bold: cases underwent infliximab treatment.
Abbreviations: No., number of cases reported in the reference; F, female; M, male; Age, age at diagnosis in years; CD, Crohn's disease; UC, ulcerative colitis; IBD, inflammatory bowel disease; CRMO, chronic recurrent multifocal osteomyelitis; Time of IBD onset, onset of IBD symptoms from occurrence of CRMO symptoms; NSAID, non-steroidal anti-inflammatory drug; 5-ASA, 5-aminosalicilate; IFX, infliximab; MTX, methotrexate; 6-MP, 6-mercaptopurine; EEN, exclusive enteral nutrition; NA, not applicable, indicating no information in the published literature.

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innate immune system, including neutrophils, macrophages, monocytes, and associated cytokines, contribute to disease pathogenesis. Increased proinflammatory cytokines, such as tumor necrosis factor (TNF)-α and interleukin (IL)-6, and decreased anti-inflammatory cytokines (especially IL-10) were reported in children with CRMO.[22] Although no definite disease-causing mutations have been identified, the increased incidence of CRMO and other inflammatory diseases in first-degree relatives of patients with CRMO suggests a significant genetic contribution.[11] For the clinician, it is essential to remember that CRMO symptoms precede IBD symptoms in 3% to 7% of CRMO patients.[23] In our patient, bone disease proceeded IBD symptoms by about two years. However, as shown in **Table 1**, IBD and CRMO can present concurrently in several cases.

NSAIDs, including naproxen (most common), indomethacin, or meloxicam, often are used as the first-line treatment for children with CRMO. Based on the study of Morbach et al.,[14] responders have significant pain relief and a decrease in the number of bone lesions on MRI by as early as three months. Patients who have persistent bone pain and hyperintense signals within the bone marrow on imaging studies after three months of NSAID treatment are considered NSAID treatment failures. These patients require a second-line treatment such as bisphosphonates, glucocorticoids, pamidronate, or biological agents including infliximab. Infliximab is a chimeric monoclonal immunoglobulin G1k antibody directed against TNF-α and inactivates TNF biologically, leading to a decrease in a large number of cytokines. Infliximab has proven effective in both the acute management of CD and as a maintenance therapy, as well as working on metastatic CD and CD-associated spondyloarthropathy, pancreatitis, pulmonary disease, ocular disease (uveitis), and cutaneous disease (psoriasis, pyoderma gangrenosum, and hidradenitis suppurativa). In several cases, infliximab was effective in CRMO with CD not only in intestinal symptoms, but also in joint symptoms.[15] Infliximab treatment was effective in our patient, supporting it as a promising third-line treatment for CRMO.

In conclusion, we report a patient with CRMO concomitant with CD. Infliximab treatment can be an additional practical choice for gastrointestinal symptoms and the manifestations of CRMO when first-line treatment fails.

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요약

Chronic recurrent multifocal osteomyelitis (CRMO)는 소아에서 호발하는 비감염성 염증성 골질환으로 염증성 장질환, 건선, 베게너 육아종, SAPHO 증후군 등의 다른 자가 면역 질환이 병발하기도 한다. 13세 중앙아시아 인종의 남자 환자가 18개월 동안 반복되는 하지 관절 통증과 연조직 염증 증상을 주소로 내원하여 CRMO로 진단 하 비스테로이드성 소염제 치료를 시작하였다. 5개월 간 약물 복용하였으나 근골격계 증상 악화, 새롭게 발생한 위장관 증상에 대해 CRMO에 크론병이 병발한 것으로 진단되었으며, 이에 대해 infliximab을 투여하였다. 본 증례에서는 크론병이 병발한 CRMO 환자에서 비스테로이드성 소염제 치료에 실패한 후 infliximab을 이용하여 효과적으로 치료된 증례를 보고하는 바이다.