

Letter to the Editor



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Incidentally Detected Asymptomatic Perianal Abscess in an Adolescent during Crohn's Disease Diagnosis: Is Routine Pelvic Imaging Required in Korean Pediatric Patients at Diagnosis?

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ABSTRACT

Perianal fistulizing diseases, namely perianal fistulas and/or abscesses, are well-known complications of Crohn's disease (CD). These are known to develop more frequently in Asian children with CD, especially in the early stages of the disease course. Approximately half of the pediatric CD cases in Korea present with perianal fistulizing diseases at diagnosis. We report a rare case of a 12-year-old boy with CD with an incidental discovery of a perianal abscess on pelvic magnetic resonance imaging during CD diagnosis. No symptoms or signs of perianal fistulizing disease were identified. The early diagnosis of the perianal abscess enabled timely and effective treatment. Considering the high incidence of concomitant perianal CD in Korean children at diagnosis, perianal imaging may be useful and should be considered during diagnostic evaluation, even in patients with no subjective or objective findings indicating perianal CD.

Keywords: Crohn disease; Abscess; Fistula; Magnetic resonance imaging

INTRODUCTION

Crohn's disease (CD) is an inflammatory bowel disease that is characterized by transmural inflammation and ulcers that develop anywhere throughout the gastrointestinal (GI) tract [1]. Nearly 25% of patients with CD are diagnosed before the age of 20 years, and its incidence has notably increased in children [1-4]. CD has a more aggressive disease course in children than in adults and requires the early introduction of immunomodulators and/or anti-tumor necrosis factor agents [5,6].

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Conflict of Interest

The authors have no financial conflicts of interest.

The well-known complications of CD include intestinal fibrosis and fistulas, which occur as a result of accumulated inflammation in the bowel [7], and perianal fistulas and/or abscesses [7]. These perianal fistulizing diseases are known to develop more frequently in pediatric CD patients of Asian ethnicity, especially at the early stages of the disease course [8]. Recent studies have also shown that the prevalence of perianal fistulizing diseases is significantly higher in Korean and Japanese children compared to their European counterparts [9-11].

When CD is diagnosed in children, a full evaluation of the entire GI tract is required to assess the extent of CD involvement to accurately designate the phenotype of disease according to the Paris classification [12]. The modalities utilized typically include ileocolonoscopy for the evaluation of the lower GI tract; esophagogastroduodenoscopy for the proximal upper GI tract; and magnetic resonance enterography (MRE), intestinal ultrasonography, and/or small bowel capsule endoscopy for the distal upper GI tract. Perianal imaging during the diagnosis of CD is indicated when patients present with symptoms and signs of perianal fistulizing diseases, and it is usually conducted using pelvic magnetic resonance imaging (MRI) or transrectal ultrasound (TRUS).

Here, we report a rare case of a 12-year-old boy with CD with a perianal abscess that was incidentally discovered at diagnosis by pelvic MRI, which was conducted as an adjunct to MRE.

CASE REPORT

A 12-year-old boy was admitted for prolonged intermittent fever, intermittent abdominal pain, and diarrhea for 3 months. The patient also experienced a weight loss of 3 kg per month, without a decrease in growth velocity. His personal medical and family histories were unremarkable.

On admission, his body temperature was 37.3°C, and all other vital signs were within the normal range. On physical examination, abdominal tenderness was absent, and there were no abnormal findings in the perianal area (**Fig. 1A**). The patient's Tanner stage was 2. The initial laboratory tests revealed the following: white blood cell count of 7,960/ μ L, erythrocyte sedimentation rate of 50 mm/h, C-reactive protein of 1.75 mg/dL, and serum albumin of 4.0 mg/dL. The patient's stool occult blood test was positive, and fecal calprotectin level was 1,326.8 mg/kg. No pathogens were detected in the stool polymerase chain reaction. Chest radiography showed no abnormal findings in the lungs, and the interferon-gamma release assay was negative.

Ileocolonoscopy revealed aphthous ulcers in the terminal ileum and colon (**Fig. 1B**), and cryptitis and crypt abscess were noted on histology. Esophagogastroduodenoscopy was unremarkable. MRE revealed multifocal asymmetric wall thickening with enhancement and diffuse restriction in the pelvic ileal loop (**Fig. 1C**). Despite the absence of symptoms or signs indicating perianal fistulizing disease, pelvic MRI was conducted as an adjunct exam to MRE, according to the protocol of the Kyungpook National University Children's Hospital. Surprisingly, a complex perianal fistula with abscess formation in the anterolateral aspect of the upper anus was detected on pelvic MRI (**Fig. 1D**). The patient was diagnosed with CD presenting as with the phenotype of A1a, L3+L4b, B1p, and G0, according to the Paris classification. Moreover, the pediatric CD activity index score was 32.5.

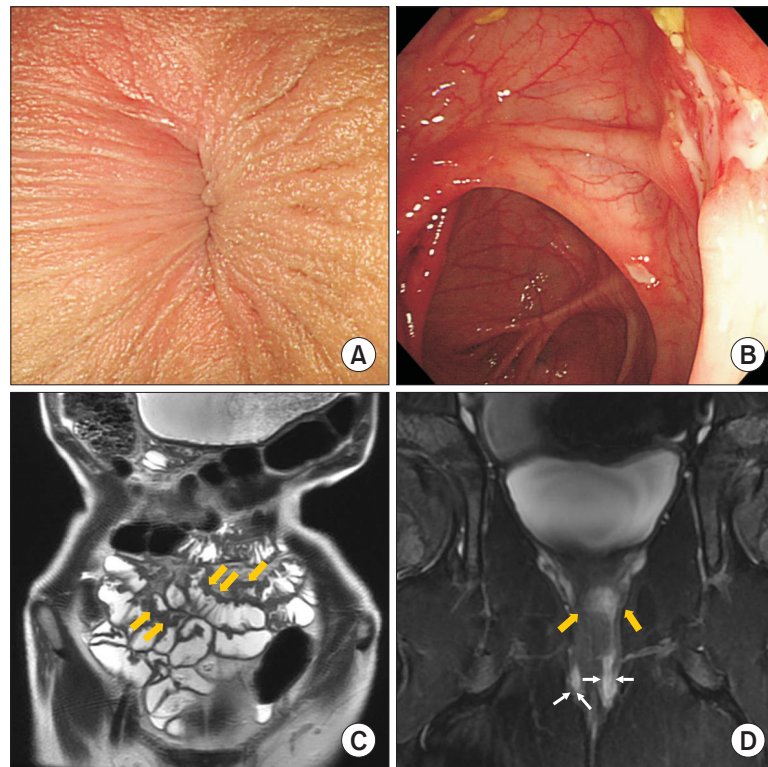


Fig. 1. At baseline evaluation of Crohn's disease. (A) No abnormal findings were noted during the inspection of the anus. (B) Ileocolonoscopy showed multifocal aphthous ulcers in the ileum and colon. (C) Magnetic resonance enterography showed multifocal asymmetric wall thickening with enhancement and diffuse restriction in the pelvic ileal loop (yellow arrows). (D) Fat-suppressed T2-weighted pelvic magnetic resonance imaging reveals a supralelevator abscess located between the urethra and the anal canal (yellow arrows) and intersphincteric fistula confined by the external sphincter (white arrows).

Inspection of the anus under general anesthesia revealed an internal opening in the anal canal (**Fig. 2A**). As such, a Seton operation was conducted (**Fig. 2B**), and azathioprine and metronidazole were initiated. One week later, the surgical wound showed delayed healing, and infliximab was then administered, considering the severity and complexity of his perianal disease. After 14 weeks, ileocolonoscopy revealed endoscopic remission and a pseudopolyp in the anal canal (**Fig. 3A**). Pelvic MRI at one-year follow-up showed complete healing of the perianal fistulizing disease (**Fig. 3B**).

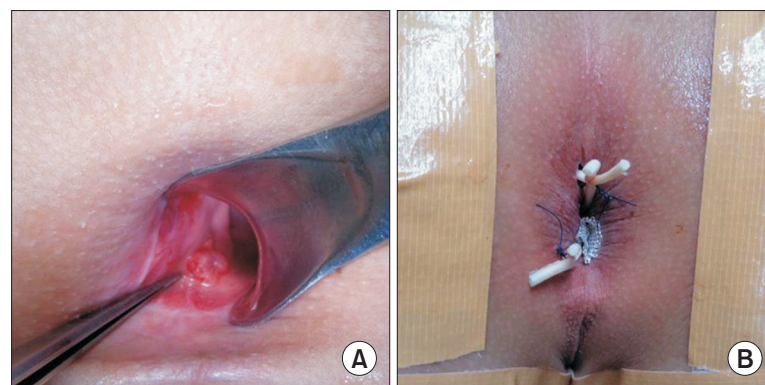


Fig. 2. Inspection of the anus under general anesthesia and seton operation. (A) Inspection of the anus under general anesthesia reveals an internal opening in the anal canal. (B) Incision, drainage, and seton placement were conducted.

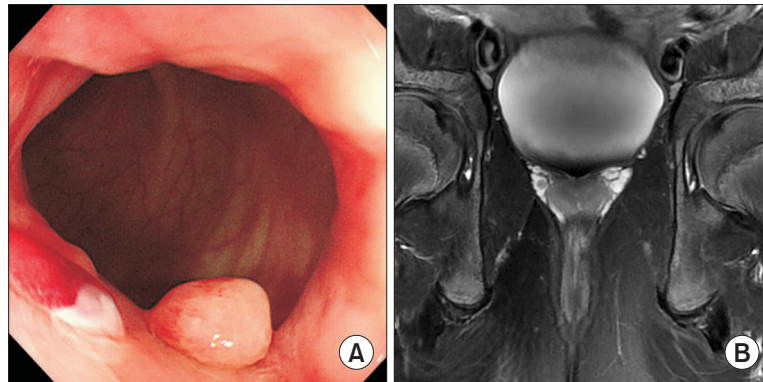


Fig. 3. At follow-up. (A) Infliximab was started one week post-operation, and ileocolonoscopy conducted after 14 weeks shows a pseudopolyp adjacent to a healing mucosa in the anal canal. (B) The pelvic magnetic resonance imaging at 1-year follow-up shows no remaining perianal abscess, perianal fistula tracts, and inflammation.

Ethics statement

This study was conducted with approval from the Institutional Review Board of Kyungpook National University Chilgok Hospital (IRB number: 2020-11-002) and in accordance with the Declaration of Helsinki (as revised in 2013).

DISCUSSION

In this case, we observed that perianal abscesses can coexist with luminal CD at diagnosis even in the absence of symptoms or signs indicating perianal CD involvement.

Perianal fistulizing diseases mostly present with the symptoms of discharge, bleeding, pain, and swelling from one or more cutaneous openings [13]. Although it is generally known that perianal fistulizing diseases develop as a complication during the natural disease course of CD, they may also be the first presentation of CD in a healthy individual. Therefore, recent guidelines recommend that all patients with unexplained perianal abscesses or complex fistulae should be investigated for luminal CD [14].

Meanwhile, radiologic examination using either pelvic MRI or TRUS is not mandatory for the diagnostic evaluation of CD. Although recent guidelines recommend that a thorough baseline clinical examination of the perianal area should be conducted in all patients newly diagnosed with CD, pelvic MRI or TRUS is only recommended for patients with symptoms and signs suspicious of active fistulizing disease to detect and delineate the anatomy of the fistula [13,14].

Perianal fistulizing diseases tend to develop more frequently in patients of Asian descent than in Caucasians during the disease course of CD [15]. Perianal fistula and/or abscess have been reported in 45-50% of Korean children with CD at diagnosis [9,16], and these rates are even higher in patients with moderate-to-severe disease activity at diagnosis [5,17-19]. Meanwhile, the incidence of perianal fistulizing disease at diagnosis in children with CD from Western countries is relatively low (4-45%) [20]. Ethnic differences may result in variations in the timing of perianal fistulizing disease development during the natural disease course of CD. Recent data have shown that perianal fistulizing diseases are likely to develop earlier in the disease course in Asian children aged ≥ 10 years compared to their Caucasian counterparts

[8]. Therefore, there may be unrevealed cases similar to the patient presented in this case study with an asymptomatic perianal fistulizing disease, especially in Korean children.

There is a scarcity of data in the literature regarding the significance of asymptomatic perianal fistulizing diseases in CD patients with asymptomatic perianal fistulas and/or abscesses. However, a recent prospective study in Korean adults with CD who underwent anal MRI supplementary to MRE at baseline diagnostic evaluation revealed that asymptomatic perianal tracts were present in 12% (53/440) of the patients [21]. The asymptomatic tracts in that study were all perianal fistulas, mostly single unbranched (83%), inter-sphincteric (72%), or with a linear dark signal at the tract margin (79%), not indicative of surgery. No cases of abscesses similar to this case were observed in that study. Meanwhile, the authors also reported that younger age, female sex, and CD activity index scores of 220-450 were independently associated with the detection of these perianal tracts. Furthermore, the detection of asymptomatic tracts on MRI was independently associated with the subsequent development of perianal fistulas or abscesses that required treatment, with an adjusted hazard ratio of 3.06 (95% confidence interval, 1.01-9.27; $p=0.048$). These results suggest that a pelvic MRI adjunct to MRE may play a crucial role in the early detection of CD patients who are at risk of perianal complications, which can help direct more attention to their management.

It is known that the presence of undiagnosed fistulas and abscesses is a major cause of disease relapse after attempted surgery [14,22]. Furthermore, accurate information regarding the presence and extent of complex perianal fistulas is required for timely and efficient medical treatment, particularly with anti-tumor necrosis factor agents, especially in children [14]. Moreover, because CD is generally accepted to be more severe and aggressive in children than in adults, precise phenotyping at diagnosis according to the Paris classification may be the first step for the precise stratification of patients that are likely to require early treatment with biologics in the treat-to-target era [6,23].

To minimize the time-consuming scanning processes and patient inconvenience, several centers in Korea, including ours, are currently performing pelvic MRI as an adjunct to regular MRE in clinical practice. Studies have shown that this approach is feasible and sensitive in detecting major perianal lesions [21,24]. A study on children with perianal CD has recently reported that MRE is capable of detecting perianal diseases with high specificity and moderate sensitivity in pediatric inflammatory bowel disease patients when using a standard technique [24]. Considering its feasibility and accuracy, along with the high incidence of perianal fistulizing disease in children and adolescents in Korea, a pelvic MRI adjunct to MRE could be considered as a routine examination at diagnosis. However, future longitudinal studies and cost-effectiveness studies are required to determine its necessity.

In conclusion, we reported a rare case of a 12-year-old boy with CD in whom a perianal abscess was incidentally discovered at diagnosis by pelvic MRI as an adjunct to MRE. The early diagnosis of perianal abscess enabled timely and effective treatment with infliximab. Considering the high incidence of concomitant perianal fistulizing diseases in Korean children at diagnosis, perianal imaging may be useful and should be considered during diagnostic evaluation, even in patients with no symptoms or signs of perianal involvement of CD.

REFERENCES

1. Rosen MJ, Dhawan A, Saeed SA. Inflammatory bowel disease in children and adolescents. *JAMA Pediatr* 2015;169:1053-60.
[PUBMED](#) | [CROSSREF](#)
2. Sýkora J, Pomahačová R, Kreslová M, Cvalínová D, Štych P, Schwarz J. Current global trends in the incidence of pediatric-onset inflammatory bowel disease. *World J Gastroenterol* 2018;24:2741-63.
[PUBMED](#) | [CROSSREF](#)
3. Roberts SE, Thorne K, Thapar N, Broekaert I, Benninga MA, Dolinsek J, et al. A systematic review and meta-analysis of paediatric inflammatory bowel disease incidence and prevalence across Europe. *J Crohns Colitis* 2020;14:1119-48.
[PUBMED](#) | [CROSSREF](#)
4. Hong SJ, Cho SM, Choe BH, Jang HJ, Choi KH, Kang B, et al. Characteristics and incidence trends for pediatric inflammatory bowel disease in Daegu-Kyungpook province in Korea: a multi-center study. *J Korean Med Sci* 2018;33:e132.
[PUBMED](#) | [CROSSREF](#)
5. Kang B, Choi SY, Kim HS, Kim K, Lee YM, Choe YH. Mucosal healing in paediatric patients with moderate-to-severe luminal Crohn's disease under combined immunosuppression: escalation versus early treatment. *J Crohns Colitis* 2016;10:1279-86.
[PUBMED](#) | [CROSSREF](#)
6. Kang B, Choe YH. Early biologic treatment in pediatric Crohn's disease: catching the therapeutic window of opportunity in early disease by treat-to-target. *Pediatr Gastroenterol Hepatol Nutr* 2018;21:1-11.
[PUBMED](#) | [CROSSREF](#)
7. Kim S. Surgery in pediatric Crohn's Disease: indications, timing and post-operative management. *Pediatr Gastroenterol Hepatol Nutr* 2017;20:14-21.
[PUBMED](#) | [CROSSREF](#)
8. Adler J, Dong S, Eder SJ, Dombkowski KJ. ImproveCareNow Pediatric IBD Learning Health System. Perianal Crohn disease in a large multicenter pediatric collaborative. *J Pediatr Gastroenterol Nutr* 2017;64:e117-24.
[PUBMED](#) | [CROSSREF](#)
9. Kang B, Kim JE, Jung JH, Choe JY, Kim MJ, Choe YH, et al. Korean children and adolescents with Crohn's disease are more likely to present with perianal fistulizing disease at diagnosis compared to their European counterparts. *Pediatr Gastroenterol Hepatol Nutr* 2020;23:49-62.
[PUBMED](#) | [CROSSREF](#)
10. Arai K, Kunisaki R, Kakuta F, Hagiwara SI, Murakoshi T, Yanagi T, et al. Phenotypic characteristics of pediatric inflammatory bowel disease in Japan: results from a multicenter registry. *Intest Res* 2020;18:412-20.
[PUBMED](#) | [CROSSREF](#)
11. de Bie CI, Paerregaard A, Kolacek S, Ruemmele FM, Koletzko S, Fell JM, et al. Disease phenotype at diagnosis in pediatric Crohn's disease: 5-year analyses of the EUOKIDS Registry. *Inflamm Bowel Dis* 2013;19:378-85.
[PUBMED](#) | [CROSSREF](#)
12. Levine A, Griffiths A, Markowitz J, Wilson DC, Turner D, Russell RK, et al. Pediatric modification of the Montreal classification for inflammatory bowel disease: the Paris classification. *Inflamm Bowel Dis* 2011;17:1314-21.
[PUBMED](#) | [CROSSREF](#)
13. Steinhart AH, Panaccione R, Targownik L, Bressler B, Khanna R, Marshall JK, et al. Clinical practice guideline for the medical management of perianal fistulizing Crohn's disease: the Toronto consensus. *Inflamm Bowel Dis* 2019;25:1-13.
[PUBMED](#) | [CROSSREF](#)
14. Maaser C, Sturm A, Vavricka SR, Kucharzik T, Fiorino G, Annese V, et al. ECCO-ESGAR Guideline for Diagnostic Assessment in IBD Part 1: initial diagnosis, monitoring of known IBD, detection of complications. *J Crohns Colitis* 2019;13:144-64.
[PUBMED](#) | [CROSSREF](#)
15. Shi HY, Levy AN, Trivedi HD, Chan FKL, Ng SC, Ananthkrishnan AN. Ethnicity influences phenotype and outcomes in inflammatory bowel disease: a systematic review and meta-analysis of population-based studies. *Clin Gastroenterol Hepatol* 2018;16:190-7.e11.
[PUBMED](#) | [CROSSREF](#)

16. Kim BJ, Song SM, Kim KM, Lee YJ, Rhee KW, Jang JY, et al. Characteristics and trends in the incidence of inflammatory bowel disease in Korean children: a single-center experience. *Dig Dis Sci* 2010;55:1989-95.
[PUBMED](#) | [CROSSREF](#)
17. Kang B, Lee Y, Lee K, Choi YO, Choe YH. Long-term outcomes after switching to CT-P13 in pediatric-onset inflammatory bowel disease: a single-center prospective observational study. *Inflamm Bowel Dis* 2018;24:607-16.
[PUBMED](#) | [CROSSREF](#)
18. Kang B, Choi SY, Choi YO, Kim MJ, Kim K, Lee JH, et al. Subtherapeutic infliximab trough levels and complete mucosal healing are associated with sustained clinical remission after infliximab cessation in paediatric-onset Crohn's disease patients treated with combined immunosuppressive therapy. *J Crohns Colitis* 2018;12:644-52.
[PUBMED](#) | [CROSSREF](#)
19. Kang B, Choi SY, Choi YO, Lee SY, Baek SY, Sohn I, et al. Infliximab trough levels are associated with mucosal healing during maintenance treatment with infliximab in paediatric Crohn's disease. *J Crohns Colitis* 2019;13:189-97.
[PUBMED](#) | [CROSSREF](#)
20. Duricova D, Fumery M, Annese V, Lakatos PL, Peyrin-Biroulet L, Gower-Rousseau C. The natural history of Crohn's disease in children: a review of population-based studies. *Eur J Gastroenterol Hepatol* 2017;29:125-34.
[PUBMED](#) | [CROSSREF](#)
21. Kim PH, Park SH, Jin K, Ye BD, Yoon YS, Lee JS, et al. Supplementary anal imaging by magnetic resonance enterography in patients with Crohn's disease not suspected of having perianal fistulas. *Clin Gastroenterol Hepatol* 2020;18:415-23.e4.
[PUBMED](#) | [CROSSREF](#)
22. Buchanan G, Halligan S, Williams A, Cohen CR, Tarroni D, Phillips RK, et al. Effect of MRI on clinical outcome of recurrent fistula-in-ano. *Lancet* 2002;360:1661-2.
[PUBMED](#) | [CROSSREF](#)
23. Bouguen G, Levesque BG, Feagan BG, Kavanaugh A, Peyrin-Biroulet L, Colombel JF, et al. Treat to target: a proposed new paradigm for the management of Crohn's disease. *Clin Gastroenterol Hepatol* 2015;13:1042-50.e2.
[PUBMED](#) | [CROSSREF](#)
24. AlSabban Z, Carman N, Moineddin R, Lo RT, King SK, Langer JC, et al. Can MR enterography screen for perianal disease in pediatric inflammatory bowel disease? *J Magn Reson Imaging* 2018;47:1638-45.
[PUBMED](#) | [CROSSREF](#)