MAYO CLINIC PROCEEDINGS: INNOVATIONS, QUALITY & OUTCOMES

CASE REPORT

Severe Alveolar Bone Loss as a Rare Manifestation of Suspected Intestinal Behçet Disease: A Case Report

Satoru Morikawa, DDS, PhD; Yuta Kaieda, MD; Satoshi Usuda, DDS; Kazuyuki Tsunoda, DDS, PhD; Takanori Kanai, MD, PhD; and Taneaki Nakagawa, DDS, PhD

Abstract

Oral ulcers commonly occur in Behçet disease, but severe alveolar bone loss is rarely documented. We report a case where marked periodontal destruction preceded the diagnosis of suspected intestinal Behçet disease. A 21-year-old man presented with extensive palatal ulcers and severe maxillary alveolar bone loss extending to the root apices. Radiographic examination revealed localized bone destruction confined to the palatal region. Subsequent evaluation revealed intestinal lesions characteristic of Behçet disease. A combination of systemic medications (prednisolone, mesalazine, and infliximab) and periodontal care stabilized both oral and intestinal conditions. This case highlights that severe alveolar bone destruction can manifest as a complication of intestinal Behçet disease, underscoring the importance of thorough oral examinations and integrated medical-dental care.

Behçet disease is a chronic multisystem inflammatory disorder characterized by recurrent oral ulcers, genital ulcers, ocular inflammation, and skin manifestations.[1] It occurs with notably higher frequencies in the Eastern Mediterranean and East Asian regions, with reported prevalence rates ranging between 13 and 20 per 100,000 individuals in Japan and 400 per 100,000 individuals in Turkey.[1]

Gastrointestinal involvement, occurring in approximately 3% to 60% of patients, represents a distinct subset known as intestinal Behçet disease and can substantially impact patient outcomes.[2,3] Although recurrent oral aphthous ulcers are a cardinal manifestation and major diagnostic criterion of Behçet disease, occurring in 95% to 100% of patients,[2] the spectrum of oral involvement often extends beyond typical ulcerative lesions. Some patients develop severe periodontal inflammation and tissue destruction, suggesting a broader impact on oral health.[4] However, the relationship between periodontal breakdown and systemic manifestations, particularly in intestinal Behçet disease, remains poorly understood.[5] The heterogeneity of oral manifestations poses substantial diagnostic challenges, especially when severe periodontal destruction precedes other systemic symptoms.

In this report, we present a case of suspected intestinal Behçet disease where severe localized alveolar bone loss preceded the diagnosis of intestinal involvement. This unusual presentation highlights 2 critical aspects: the potential for severe periodontal destruction as an early manifestation of systemic disease and the importance of collaborative medical-dental care in diagnosis and management. The case can expand our understanding of the oral manifestations in Behçet disease and underscores the importance of comprehensive oral examination in disease surveillance, which may facilitate earlier diagnosis and improved patient outcomes.

From the Department of Dentistry and Oral Surgery (S.M., S.U., K.T., T.N.), and Division of Gastroenterology and Hepatology, Department of Internal Medicine (Y.K., T.K.), Keio University School of Medicine, Shinjuku-ku, Tokyo, Japan.

CASE PRESENTATION

A 21-year-old man presented with multiple, painful palatal ulcers, tooth mobility, and recurrent nonbloody loose stools. He had no

ocular or joint involvement. He had a white, spotted pubic rash that resolved 2 years before presentation. On the basis of its clinical presentation, this rash does not fully align with the dermatological criteria outlined in the International Criteria for Behçet disease (ICBD; 2014), which include papulopustular lesions, pseudofolliculitis, erythema nodosum, and superficial thrombophlebitis. However, it may still represent a nonclassical cutaneous manifestation of Behçet disease. Six months before presentation, he developed a painful palatal gingival ulcer unresponsive to antibacterial treatment. Worsening symptoms prompted referral to oral surgery and periodontics, where necrotizing periodontitis was suspected.

The medical history and blood test results at that juncture were unremarkable. Physical examination showed palatal pseudomembranous ulcerations from the right to left maxillary second molars with pronounced gingival recession (Figures 1A-C). Cone-beam computed tomography revealed severe localized alveolar bone resorption from the right to left maxillary second molars, with bone loss extending to the root apices in the axial view (Figure 1D) and sagittal views of the right (Figure 1E) and left (Figure 1F) sides. No dermatologic manifestations were evident.

Despite no abnormal findings on esophagogastroduodenoscopy, colonoscopy revealed an oval-shaped cecal erosion (Figure 2A) and chronic lower rectal inflammation (Figure 2B) characterized by fine granular and nodular mucosae resembling lymphoid follicular proctitis. Histopathologic examination revealed cryptitis. No granulomas were detected in the cecum. We considered that he also had previous episodes of recurrent stomatitis and pubic rash, manifestations of the ICBD, and the oval-shaped cecal erosion could be endoscopic findings of intestinal Behçet disease. Indeed, intestinal Behçet disease usually affect ileocecal mucosa and has round erosions.[6] Hence, intestinal Behçet disease was suspected.

Prednisolone was initiated, with considerable improvement within a month. Subsequently, mesalazine (3 g) and colchicine (0.5 mg) were introduced with ongoing endoscopic evaluation. Tapering prednisolone resulted in worsening of the intestinal ulceration, as noted on colonoscopy. Oval-

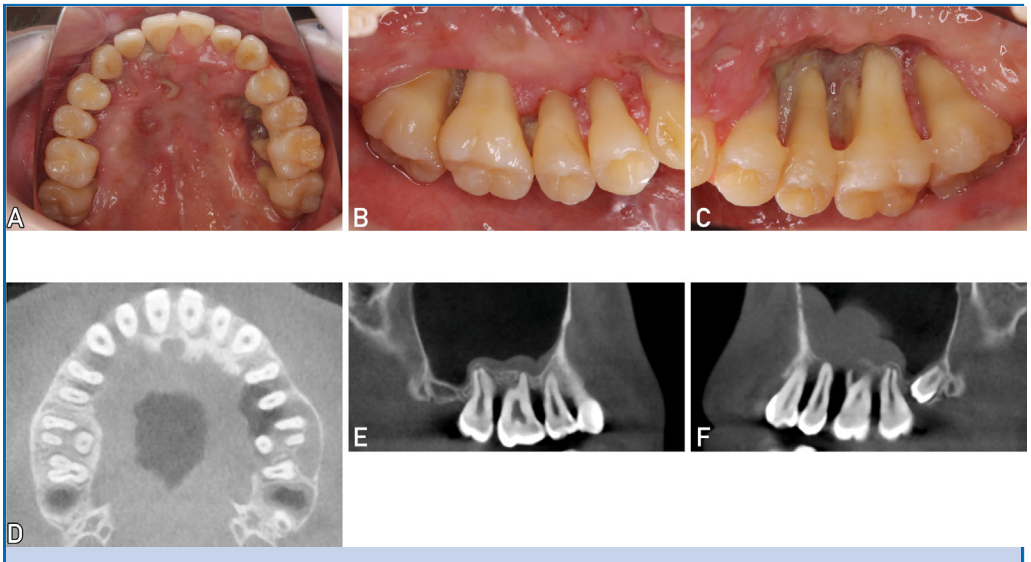

FIGURE 1. Initial oral examination and radiographic findings of maxillary alveolar bone resorption. (A-C) Intraoral photographs showing extensive palatal aphthae with severe root exposure and alveolar bone resorption confined to the palatal region: occlusal view (A), right palatal view (B), and left palatal view (C). (D-F) Cone-beam computed tomography images demonstrating severe alveolar bone loss: axial view showing extensive bone resorption in the bilateral posterior regions (D), and sagittal views of the right (E) and left (F) molar regions showing bone loss extending to the root apices.

Mayo Clin Proc Inn Qual Out ■ June 2025;9(3):100612 ■ https://doi.org/10.1016/j.mayocpiqo.2025.100612
www.mcpiqojournal.org

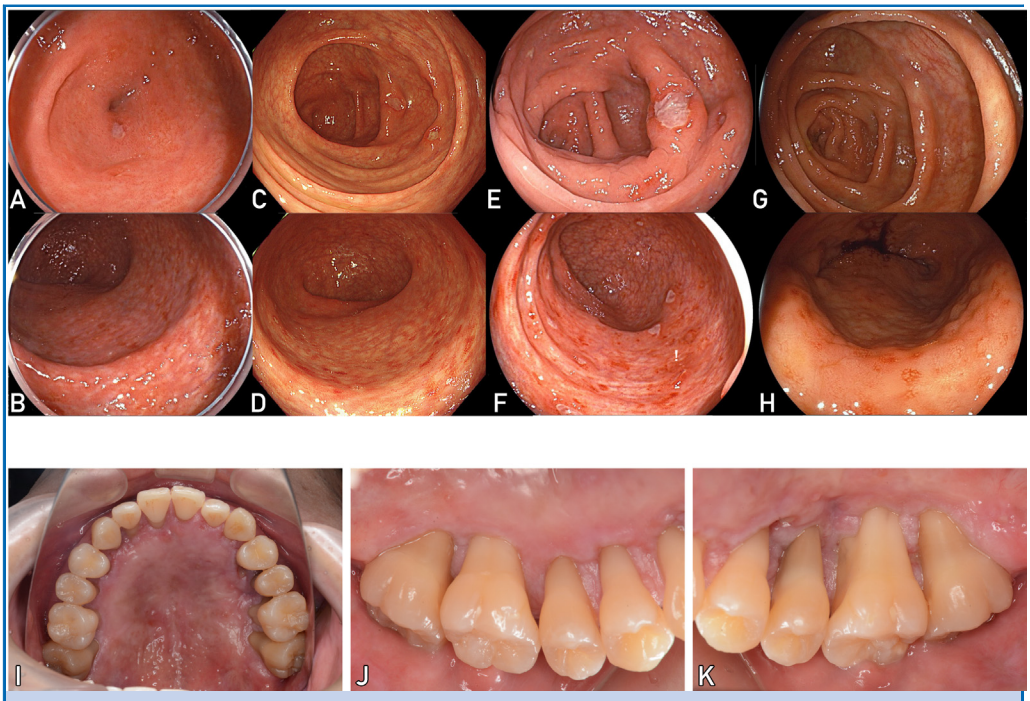

FIGURE 2. Disease progression and treatment response. (A-H) Endoscopic images of the gastrointestinal tract at various stages of treatment—(A, B) initial consultation: (A) oval-shaped cecal erosion and (B) chronic lower rectal inflammation; (C, D) after prednisolone therapy: (C) multifocal oval-shaped erosions in the ascending colon and (D) chronic lower rectal inflammation with subtle erosions; (E, F) After azathioprine treatment: (E) exacerbation of ulcers in the ascending colon and (F) development of oval-shaped erosions in the lower rectum; and (G, H) After infliximab therapy: (G) resolution of ulcers in the ascending colon and (H) significant improvement in oval-shaped ulcers in the lower rectum. (I-K) Posttreatment oral findings demonstrating improvements in ulceration: occlusal view showing epithelial regeneration in the previously ulcerated palatal region (I), right palatal view (J), and left palatal view (K).

shaped ulcers (Figure 2C), typical of intestinal Behçet disease, were detected after negative cytomegalovirus staining. Further, subtle erosion was detected in the lower rectum (Figure 2D). Considering disease exacerbation after prednisolone tapering, azathioprine was administered.

Genetic testing revealed the ARG/ARG genotype of *NUDT15*, a marker associated with azathioprine sensitivity. Therefore, to address the refractory nature of the oral and intestinal symptoms, azathioprine 50 mg was initiated. After 2 months, the stools became solid. Despite oral and bowel symptom amelioration, follow-up colonoscopy showed an increase in the number and size of ulcers (Figures 2E and F). Consequently, infliximab (5 mg/kg) was infused intravenously every 8 weeks for treating intestinal Behçet disease. Follow-up colonoscopy demonstrated ulcer resolution in the ascending colon and substantial improvement of ulcers in the lower rectum (Figures 2G and H).

Further treatment included diligent periodontal care and continued pharmacotherapy. After the initiation of systemic corticosteroid therapy (prednisolone), mesalazine, and colchicine combined with comprehensive periodontal treatment, the oral ulcerations substantially improved, showing epithelial regeneration in the palatal region (Figures 2I–K). Occasional recurrence of mild localized oral ulcers was evident. The overall oral and gastrointestinal condition was stable. Follow-up dental radiographs indicated no further progression of maxillary alveolar bone resorption, suggesting periodontal condition stabilization.

Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

DISCUSSION

This case report highlighted 2 substantial findings in a patient with suspected intestinal Behçet disease: extensive alveolar bone resorption with bone loss extending to the root apices, representing a novel oral manifestation, and severe oral symptoms, including painful stomatitis, marked gingival recession, and increased tooth mobility, preceding the diagnosis of suspected intestinal lesions. This case highlights the importance of a multidisciplinary approach in managing suspected intestinal Behçet disease with severe oral manifestations. These findings suggest a potential link between severe periodontal deterioration and systemic manifestations of Behçet disease, particularly its suspected intestinal form.

The severe alveolar bone resorption observed in this case is a rarely reported oral manifestation of Behçet disease, although oral ulcers are well documented.[7] Severe and extensive alveolar bone resorption in a patient with suspected intestinal Behçet's disease has not been reported previously. This finding can expand our understanding of the potential oral complications of Behçet disease. Previous studies have primarily focused on aphthous ulcers as the primary oral manifestation. However, our case demonstrated that oral involvement may be more extensive and serious. The severity of alveolar bone loss suggested a possible connection between the inflammatory processes in Behçet disease and accelerated periodontal breakdown. However, the reason for such substantial ulceration and concentration of alveolar bone resorption solely in the palatal region remains unclear, warranting further investigation.

Our case emphasizes the potential of oral symptoms as an early indicator of suspected intestinal Behçet disease. The presence of multiple aphthae (particularly on the palate) and severe alveolar bone resorption should prompt clinicians to consider the possibility of underlying intestinal diseases, including Behçet disease, which may otherwise remain undiagnosed. This aligns with a recent study's findings suggesting a complex interplay between oral health and systemic inflammatory conditions.[4] Considering that untreated bacterial infections associated with such oral manifestations can lead to further bone resorption and subsequent tooth loss, prompt and thorough examination and treatment are important. This unique presentation highlights the need for dental professionals to remain vigilant when examining patients with suspected systemic inflammation.

Although the pubic rash in this case was a genital ulcer caused by Behçet disease, this case did not meet the 1987 Japanese diagnostic criteria for Behçet disease.[8] However, if this assumption is true, it aligned with the 1990 International Study Group criteria[9] and 2006 ICBD.[10]

Diagnostic criteria for intestinal Behçet disease define ulcers as typical if they meet all the following conditions: <5 in number, oval in shape, deep, with discrete borders, and located in the ileocecal area. Ulcers that do not meet all these conditions are classified as atypical.[6] Then, the presence of Behçet disease symptoms is used to distinguish among definite, probable, suspected, and undiagnosed Behçet disease.

When applying these diagnostic criteria to this patient, the ulcers were classified as atypical owing to their distribution. The presence of Behçet disease symptoms, including oral and pubic ulcers, led to its classification as probable intestinal Behçet disease. However, because a definitive diagnosis can only be made after observing the disease course over time, we present this case as suspected intestinal Behçet disease.

This study's clinical implications are diverse. First, this case emphasizes the importance of oral health as a key, yet often neglected, component of systemic disease surveillance, especially in the context of Behçet disease. Second, it highlights the need for a multidisciplinary approach in diagnosing and treating complex cases presenting with oral ulcerations and alveolar bone loss. These observations align with recent recommendations for the integrated care of autoimmune diseases.[11] In practice, this would involve

collaborative clinics where dentists, periodontists, rheumatologists, and gastroenterologists jointly assess patients with suspected Behçet disease. Such a strategy may facilitate earlier detection of intestinal involvement and more comprehensive management, potentially improving the long-term outcomes in these patients.

This case also has certain implications in the current classification of periodontal diseases. The 2017 World Workshop on the Classification of Periodontal and Peri-Implant Diseases and Conditions introduced a new system for categorizing periodontitis and peri-implant diseases, recognizing the potential systemic effects of periodontal diseases.[12,13] This classification also provides case definitions and diagnostic considerations for systemic diseases affecting the periodontal attachment.[14] However, Behçet disease is not currently listed among the systemic diseases directly manifesting as periodontitis. Our findings suggest that severe alveolar bone loss may be a manifestation of intestinal Behçet disease, which warrants its inclusion in this classification in future updates.

This case revealed the occurrence of severe alveolar bone resorption as a possible manifestation of intestinal Behçet disease, which is a seldom reported finding. This highlights the need for thorough oral examinations in patients with suspected Behçet disease and emphasizes the importance of a multidisciplinary approach. Dental professionals and gastroenterologists should consider intestinal Behçet disease in patients with severe periodontal symptoms and gastrointestinal issues.

Our study revealed a gap in the research on oral manifestations specific to Behçet disease, particularly regarding severe periodontal involvement. These insights could inform future clinical guidelines and improve patient outcomes through enhanced integrated health care. Our findings may also contribute to future updates of periodontal and peri-implant condition classifications.

Further research should explore the mechanisms linking intestinal inflammation and alveolar bone loss in Behçet disease, potentially leading to targeted therapies and improved outcomes. This case highlights the evolving nature of disease classification and the need for ongoing reassessments based on new evidence.

POTENTIAL COMPETING INTERESTS

The authors have no competing interests to report.

ETHICS STATEMENT

Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

ACKNOWLEDGMENTS

We thank Editage (www.editage.com) for the English language editing. Drs Morikawa and Kaieda contributed equally to this work.

Correspondence: Address to Satoru Morikawa, DDS, PhD, Department of Dentistry and Oral Surgery, Keio University School of Medicine, 35 Shinanomachi, Shinjuku-ku, Tokyo 160-8582, Japan (morikawa@keio.jp).

ORCID

Satoru Morikawa: https://orcid.org/0000-0001-8859-2888

REFERENCES

1. Dzhus MB, Karasevska TA, Tsaralunga VM, Yurchenko AV, Ivashkivsky OI. Behçet's disease with intestinal involvement: case-based review. *Rheumatol Int*. 2022;42(9):1653-1660. https://doi.org/10.1007/s00296-022-05152-x.
2. Davatchi F, Chams-Davatchi C, Shams H, et al. Behcet's disease: epidemiology, clinical manifestations, and diagnosis. *Expert Rev Clin Immunol*. 2017;13(1):57-65. https://doi.org/10.1080/1744666X.2016.1205486.
3. Skef W, Hamilton MJ, Arayssi T. Gastrointestinal Behçet's disease: a review. *World J Gastroenterol*. 2015;21(13):3801-3812. https://doi.org/10.3748/wjg.v21.i13.3801.
4. Mumcu G, Ergun T, Inanc N, et al. Oral health is impaired in Behçet's disease and is associated with disease severity. *Rheumatology (Oxford)*. 2004;43(8):1028-1033. https://doi.org/10.1093/rheumatology/keh236.
5. Mumcu G, Öner FA, Ergun T, Direskeneli RH. Decreasing incidence and severity of Behçet's disease: a changing trend in epidemiological spectrum possibly associated with oral health. *Turk J Med Sci*. 2020;50(s1-2):1587-1590. https://doi.org/10.3906/sag-2003-147.
6. Cheon JH, Kim ES, Shin SJ, et al. Development and validation of novel diagnostic criteria for intestinal Behçet's disease in Korean patients with ileocolonic ulcers. *Am J Gastroenterol*. 2009;104(10):2492-2499. https://doi.org/10.1038/ajg.2009.331.
7. Hatemi G, Seyahi E, Fresko I, Talarico R, Uçar D, Hamuryudan V. Behçet's syndrome: one year in review 2022. *Clin Exp Rheumatol*. 2022;40(8):1461-1471. https://doi.org/10.55563/clinexprheumatol/h4dkrs.
8. Mizushima Y. [Revised diagnostic criteria for Behçet's disease in 1987]. *Ryumachi*. 1988;28(1):66-70.
9. International Study Group for Behçet's Disease. Criteria for diagnosis of Behçet's disease. *Lancet*. 1990;335(8697):1078-1080. https://doi.org/10.1016/0140-6736(90)92643-V.

10. International Team for the Revision of the. International Criteria for Behcet's Disease. Evaluation of the International Criteria for Behcet's disease (ICBD). *Clin Exp Rheumatol*. 2006;24(suppl 42):S13.
11. Yazici H, Seyahi E, Hatemi G, Yazici Y. Behçet syndrome: a contemporary view. *Nat Rev Rheumatol*. 2018;14(2):107-119. https://doi.org/10.1038/nrrheum.2017.208.
12. Papapanou PN, Sanz M, Buduneli N, et al. Periodontitis: Consensus report of workgroup 2 of the 2017 World Workshop on the Classification of Periodontal and Peri-Implant Diseases and Conditions. *J Periodontol*. 2018; 89(suppl 1):S173-S182. https://doi.org/10.1002/JPER.17-0721.
13. Tonetti MS, Greenwell H, Kornman KS. Staging and grading of periodontitis: Framework and proposal of a new classification and case definition. *J Periodontol*. 2018;89(suppl 1):S159-S172. https://doi.org/10.1002/JPER.18-0006.
14. Albandarv JM, Susin C, Hughes FJ. Manifestations of systemic diseases and conditions that affect the periodontal attachment apparatus: case definitions and diagnostic considerations. *J Periodontol*. 2018;89(suppl 1):S183-S203. https://doi.org/10.1002/JPER.16-0480.