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CASE REPORT

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A rare case of bronchial stenosis in ulcerative colitis

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Abstract

Background Inflammatory bowel diseases (IBDs), including ulcerative colitis (UC), are chronic inflammatory disorders of the gastrointestinal tract with increasing incidence worldwide. Though primarily affecting the digestive system, IBD can lead to extraintestinal manifestations, including rare respiratory complications.

Case presentation In our case report, we present the case of a 56-year-old woman with a history of UC and bronchiectasis who developed bronchial stenosis, manifesting as worsening cough and bronchospasm. Initial treatment with corticosteroids and biologic therapy was unsuccessful. The patient subsequently underwent bronchoscopic dilatation interventions, improving her bronchial stenosis and respiratory symptoms. Maintenance therapy with corticosteroids was initiated, while gentamicin was locally administered during the procedures to control infection due to high-load colonization by Pseudomonas.

Conclusion This case underscores the importance of early diagnosis and a multidisciplinary treatment approach to managing complex IBD-related respiratory involvement. Future research on targeted therapies and personalized management strategies is needed to improve outcomes for patients with these rare complications.

Keywords Ulcerative colitis, Bronchoscopy, Bronchial stenosis, Ustekilumab

Background

Inflammatory bowel diseases (IBDs) are chronic gastroenteric inflammatory disorders characterized by a natural history of relapsing-remitting flares of the disease, including ulcerative colitis (UC) and Crohn's disease (CD) [1]. In the last decade, IBD incidence is rising all over the world [2]. In fact, IBDs affect around 7 million people globally, with regional variations, and significantly impact patients' lives and burdening healthcare systems economically [3]. Patients typically develop IBD between the ages of 15 and 30, with a second peak in onset between 40 and 50 [4]. Even if the aetiology of IBDs is not yet well known, different risk factors are underlined, such as host genetics, gut microbiota alterations, and environmental factors, which could contribute to chronic inflammation. In almost 50% of patients with IBD, the symptoms are not only limited to the gastrointestinal tract [5]. Patients with IBD may also experience extraintestinal manifestations such as joint, skin, eyes, and liver complications [5].

Respiratory involvement is a rare manifestation that often goes undiagnosed, leading to delays in proper treatment and a worsened prognosis for the patient. The prevalence of thoracic involvement in IBD is estimated to be

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less than 1%, though this figure may be underestimated [6, 7]. Recent studies indicate that 40–60% of patients with IBD show abnormalities on pulmonary function tests (PFTs) or chest computed tomography (CT) scans [8, 9]. The most common respiratory diseases associated with IBD are chronic bronchitis, subglottic stenosis, bronchiectasis and bronchiolitis, which can further make it difficult to recognize IBD-related respiratory issues [10]. Thoracic involvement is more commonly observed in patients with UC than in those with other extraintestinal manifestations of IBD [11]. In most cases, thoracic symptoms develop several years after digestive symptoms [12]. However, in less than 15% of patients, these thoracic symptoms may occur simultaneously with or even precede the onset of gastrointestinal manifestations [12]. Additionally, thoracic involvement has been reported in patients with UC, particularly following curative intestinal surgery [8].

Recognizing respiratory manifestations of IBD is important for early detection and treatment, which can prevent the progression of lung involvement and improve patient outcomes and quality of life. We present a rare case of bronchial stenosis due to the localization of UC in a female patient, which required a multidisciplinary management.

Case report

A 56-year-old caucasian woman affected by UC since 1988, treated with total colectomy in 2002, and bronchiectasis associated with UC since 1988, for whom she received left lower lobe lobectomy in 1991, presented in our clinic with worsening cough and purulent sputum. Therefore, she underwent a bronchoscopy, which showed diffuse inflamed and edematous mucosa with narrowing of the left main bronchus. The bronchial aspirate (BAL) was positive for *P. aeruginosa*. Thus, she was treated with meropenem, based on the antibiogram, with only partial improvement of the chronic symptoms. In the hypothesis of an infectious etiology, more invasive procedures, such as rigid bronchoscopy or biopsies, were not performed at that time. At the time, the patient was not receiving any specific treatment except for a low dose of steroids for asthma, as UC was in remission. Six months later, she was hospitalized for the persistence of symptoms. She presented apyretic and without hypoxia. The white blood cell count and C-reactive protein were normal. The chest CT showed stenosis of the left main bronchus with mucus plugs in peripheral bronchiectasis (Fig. 1B), not present in prior CT scans (Fig. 1A). A repeated bronchoscopy showed inflamed mucosa causing critical stenosis of the left main bronchus, and purulent secretions were observed (Fig. 2A). At this time, there was no evidence of infectious agents in BAL. Endobronchial biopsy revealed moderate chronic infiltrate, composed of lymphocytes and plasma cells, and acute inflammation with evidence of neutrophils granulocytes ulcerating superficial layers and squamous metaplasia (Fig. 3). Other common causes of bronchial stenosis were evaluated and ruled out, including prolonged intubation, cervical trauma, congenital anomalies, infections, neoplasms (benign or malignant), lung transplantation, exposure to toxic inhalants, radiation therapy, and autoimmune disorders such as sarcoidosis. The c-ANCA level of our patient was normal, and kidney function was normal, without signs or symptoms of other mucosal involvement. The bronchial biopsies were aspecific. At CT scan and bronchoscopy, respectively, there was no sign of pulmonary parenchymal or mediastinal infiltration or evidence of infectious agents, including mycobacteria. In agreement with the gastroenterologists, considering both the histopathological findings and the disease timeline, the bronchial stenosis was considered an active manifestation of UC. Therefore, the patient was treated with 1 mg/Kg/day of prednisone (50 mg/day) with a slow tapering, together with ustekinumab 260 mg/8 weeks. Despite the medical therapy, after six months she had no significant clinical, radiological (Fig. 1C), or endoscopic (Fig. 2B) improvement.

After multidisciplinary discussions, the decision was made to hospitalize the patient due to the worsening of her respiratory symptoms, such as dyspnea and cough. She underwent an endoscopic balloon dilation procedure using rigid bronchoscopy to dilate the stenosis and was treated with antibiotics. The following day, bronchoscopy revealed an improvement in the stenosis (Fig. 2C-D). No other procedures of rigid bronchoscopy were needed.

Over the following months, the patient was closely monitored for symptoms and potential exacerbations related to her respiratory condition. After multidisciplinary re-evaluation of the treatment plan, the decision was made to discontinue Ustekinumab and maintain a low dose dose of prednisone (10 mg/day). Although the left main bronchus remained patent, the patient experienced a series of infectious pulmonary exacerbations with a new isolation of *P. aeruginosa* (intermediate to cefepime, ceftazidime, ciprofloxacina, imipenem and piperacillina/tazobactam) at BAL. Subsequently, she underwent a carefully planned series of bronchoscopic procedures to administer endobronchial gentamicin directly to the affected areas (160 mg/every 3 days for 1 month). This targeted approach allowed for precise drug delivery to address localized infection. No drug toxicity, particularly ototoxicity and renal toxicity, was observed. This intervention resulted in an improvement in symptoms and exacerbations. The patient was re-evaluated every three months for the subsequent year, demonstrating stable respiratory status, and maintained UC

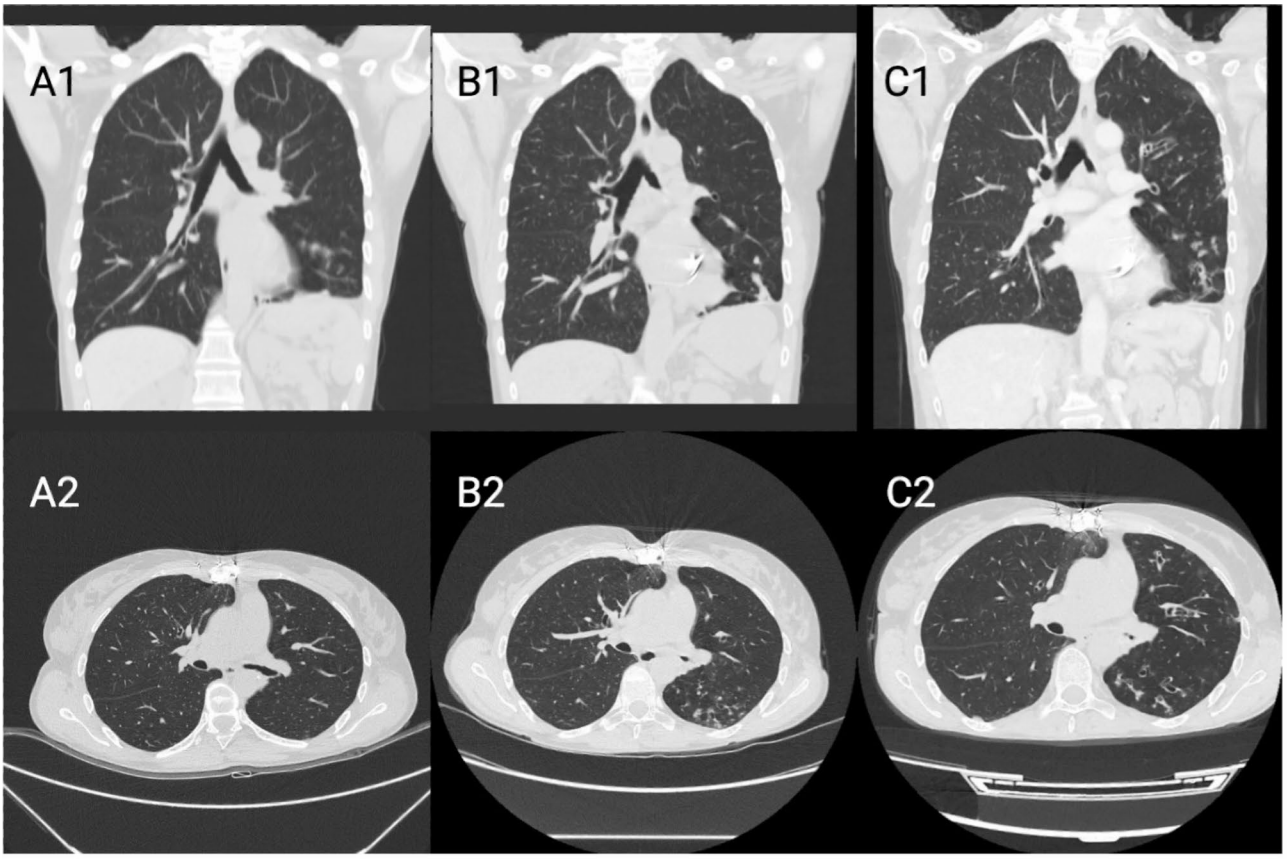

Fig. 1 (color): The coronal and axial radiological progression of stenosis of the left bronchus in a female patient at the level of carena in December 2015 (**A**), September 2022 (**B**), and February 2024 (**C**)

remission, with no significant changes in the degree of bronchial obstruction.

Discussion

Our case report showed a rare and unusual respiratory manifestation of UC in a 56-year-old Caucasian woman. After presenting with a worsening cough and purulent sputum, a bronchoscopy revealed a severe bronchial stenosis of the left main bronchus. Despite initial medical therapy, the patient's symptoms persisted, necessitating endoscopic balloon dilation.

Despite the absence of large-scale clinical trials, the literature indicates that IBD with respiratory involvement is commonly managed with corticosteroids as the primary treatment strategy [13]. Studies suggest beginning treatment with prednisone at a dose of 1 mg per kg of lean body weight per day, with a maximum limit of 60 mg per day [12]. This should be administered as a single oral dose in the morning. As the patient shows improvement, systemic glucocorticoids are tapered down and transitioned to inhaled glucocorticoids [12].

However, clinical outcomes suggest that more than 10% of patients with this condition may not achieve sufficient improvement with corticosteroid therapy alone. In cases where corticosteroids prove inadequate, immunosuppressants or monoclonal antibodies are often introduced as steroid-sparing agents or as additional treatment options, particularly in patients with UC who present with pulmonary involvement [13]. In certain rare and severe instances, it may be beneficial to consider surgical interventions such as dilation, laser therapy, or stenting to address and manage the respiratory complications associated with IBD effectively.

Prior case reports illustrate the diversity in treatment responses. For instance, Rickli and colleagues documented a case of IBD-related respiratory involvement that responded positively to steroid therapy alone, underscoring the potential efficacy of corticosteroids in some instances [14]. On the other hand, Wilcox et al. described a patient with IBD and tracheal stenosis who was initially managed with manual dilatation using a rigid bronchoscope. Unfortunately, this approach was met with complications, and the patient ultimately succumbed after a second attempt at tracheal dilatation [15]. Similarly, Plataky et al. reported the first case of a young woman with Crohn's disease who developed persistent tracheal inflammation and stenosis, which was partially responsive to corticosteroids [16]. After excluding other

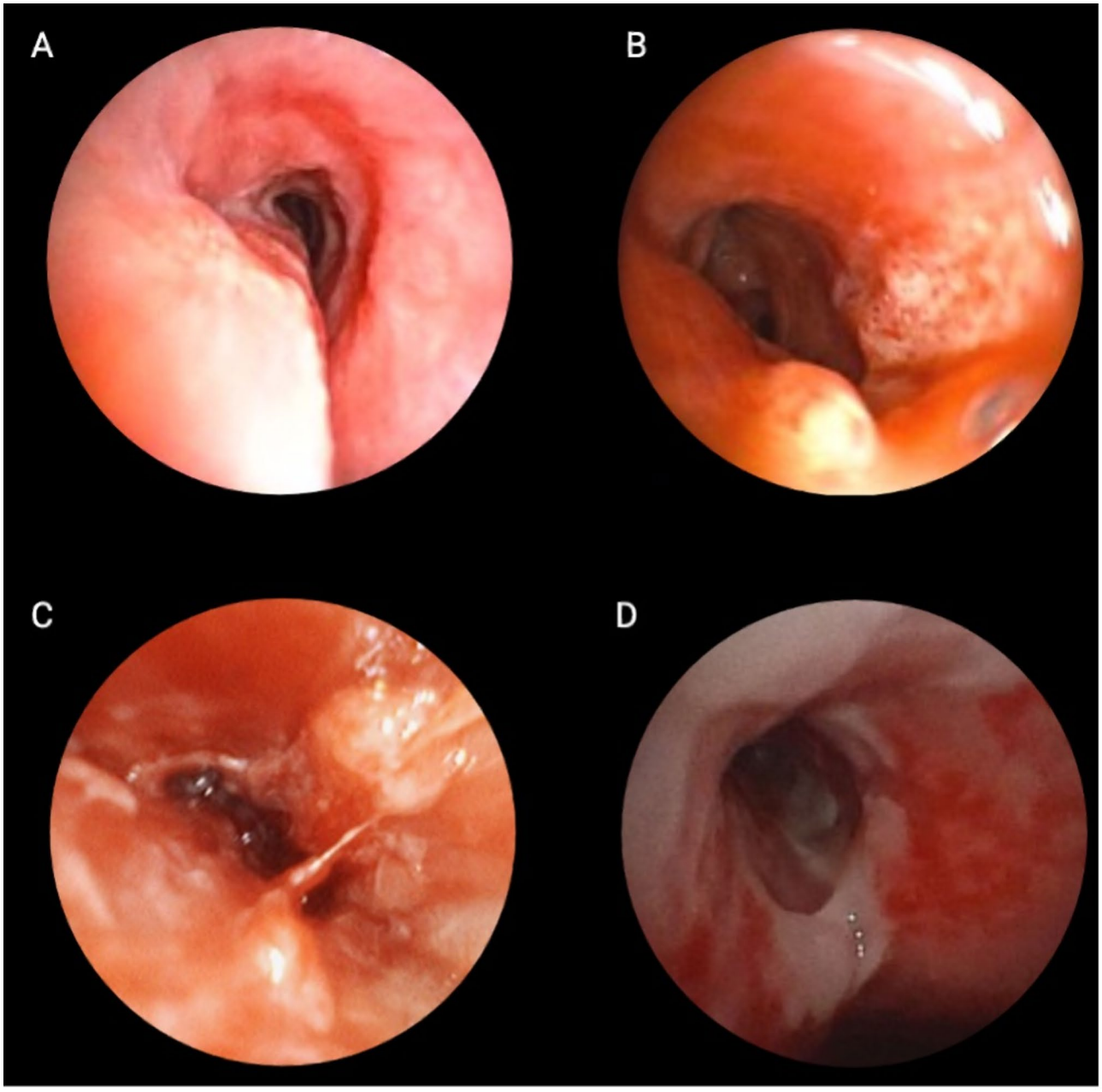

Fig. 2 (color): The endobronchial progression of the stenosis of the left bronchus: the stenosis before (**A**) and after (**B**) the pharmacology treatment; stenosis of the left bronchus before (**C**) and after (**D**) the endoscopic balloon dilation

potential causes, bronchoscopic dilation resulted in symptom remission. Finally, another case by Suzuki et al. involved a patient with bronchial stenosis associated with UC who did not respond to conventional medical therapy. This case was successfully treated with a more invasive approach, utilizing a yttrium aluminum garnet (YAG) laser to achieve improvement [17].All these cases highlight the challenges and variability in treating respiratory manifestations of IBD, emphasizing the need for individualized therapeutic strategies based on the severity and progression of the disease. Our findings show that, although rare, respiratory involvement can occur in UC and significantly impact patient outcomes if not addressed promptly. We outline an approach that includes medical therapy and interventional bronchoscopy to help clinicians manage such cases. Timely recognition and personalized care can reduce morbidity and enhance the quality of life for patients with IBD facing respiratory complications.

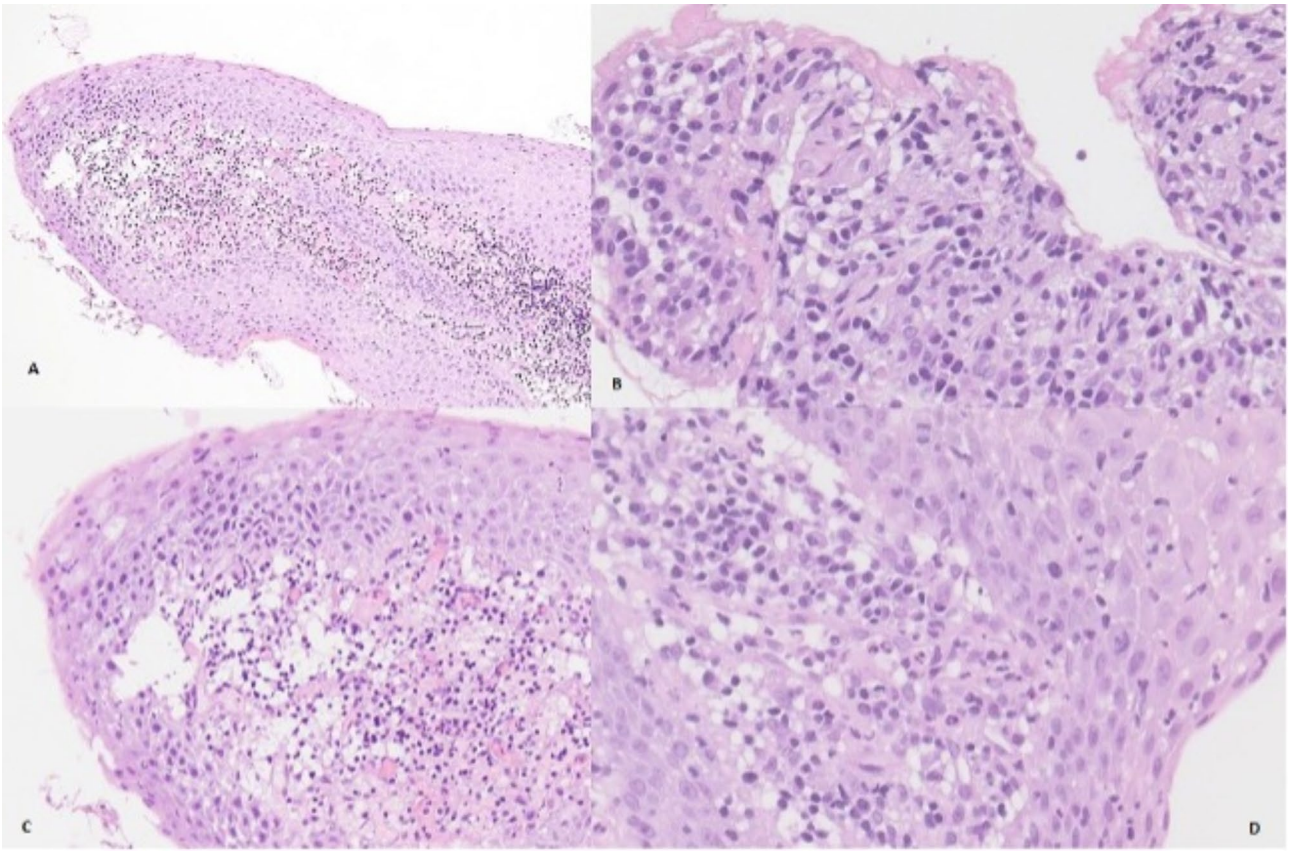

Fig. 3 (color): Endobronchial biopsy (**A**) reveals moderate chronic infiltrate, composed of lymphocytes and plasma cells (**B**), and acute inflammation with evidence of neutrophils granulocytes ulcerating superficial layers (**C**, **D**) and squamous metaplasia

Conclusion

Bronchial stenosis represents a rare but severe complication of UC [10]. Early clinical suspicion in patients with a prior history of IBD is essential to begin appropriate management. The treatment of bronchial involvement in UC is particularly challenging, as these manifestations often show limited response to conventional medical therapies and may necessitate bronchoscopic procedures or, in severe cases, surgical intervention.

Effective management of such complex cases relies on close collaboration among multiple medical specialities, including pulmonology, gastroenterology, and thoracic surgery, to provide personalized treatment for each patient's specific needs. Future perspectives include the development of new targeted therapies and personalized management strategies to improve the quality of life for patients with IBD and extra-intestinal manifestations.

Abbreviations

BAL	Bronchial aspirate
CD	Crohn's disease
CT	Computed tomography
IBD	Inflammatory bowel disease
PFT	Pulmonary function test
UC	Ulcerative colitis
YAG	Yttrium aluminum garnet

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Author contributions

Conceptualization: UZ, FA, GB, PF. Data collection: UZ, FA, AM, GP, GB, FB, FL, PF.Writing first draft: UZ, FL, PF. Critical revisions: UZ, FA, AM, GP, GB, FB, FL, PF.

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Data availability

Data available from the corresponding author upon request.

Declarations

Ethics approval and consent to participate

Not applicable.

Patient Consent for Publication

Appropriate written informed consent was obtained for publication of this case report and accompanying images.

Statement of ethics

The authors acknowledge that this research work is original and has not been previously published or is currently being considered for publication elsewhere. All sources used are properly disclosed, and all authors who have been involved are mentioned.

Competing interests

The authors declare no competing interests.

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