

# Unicentric Castleman disease with paraneoplastic pemphigus and follicular dendritic cell sarcoma: A case report

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**Abstract.** Castleman disease (CD) is a rare lymphoproliferative disorder that may be complicated by paraneoplastic pemphigus (PNP) and follicular dendritic cell sarcoma (FDCS), although their concurrence is uncommon. The present study reported on a 67-year-old woman with progressive oral ulceration and generalized purple skin spots. Imaging demonstrated a 114mm x96mm x118 mm lesion in the left pelvis. Serology was positive for anti-desmoplakin-1 and -3 and anti-BP230 antibodies. Histopathology revealed hyaline-vascular unicentric CD with focal low-to-intermediate-grade FDCS; immunohistochemistry showed CD21, CD23, CD35, CD68, CXCL13 expression and Ki-67 ~10%. Initial therapy comprised high-dose methylprednisolone, intravenous immunoglobulin and thalidomide, followed by surgical resection and adjuvant thalidomide-cyclophosphamide-prednisone. Postoperatively, inflammatory indices normalized and mucocutaneous lesions partially improved; nevertheless, the patient died of respiratory failure 6 months after surgery. In summary, the present case report aimed to provide valuable references and experiences for clinicians in the diagnosis and treatment of CD featuring PNP and FDCS.

## Introduction

Castleman disease (CD) is a rare lymphoproliferative disorder. It was first described by Benjamin Castleman in 1954 (1), with an estimated incidence of ~2 per 100,000 individuals in the US (2). CD is classified into unicentric CD (UCD) and multicentric CD (MCD) types based on the extent of lymph node

involvement. A 2024 systematic review of articles published in 1995-2021 on  $\geq 5$  cases of CD reported that UCD was slightly more frequent in women (53.7%), whereas idiopathic MCD (iMCD; 59.1%) and human herpesvirus 8 (HHV-8)+ MCD (88.5%) were predominant in men (3). The diagnosis of CD primarily relies on complete excisional biopsy of the affected lymph nodes and subsequent histological examination, with pathological features such as hyaline-vascular, plasma-cell or mixed type serving as the diagnostic gold standard. Simultaneously, it is key to exclude other diseases that can produce CD-like changes in lymph nodes (e.g., lymphoma, tuberculosis and immunological disorders) (4,5). Paraneoplastic pemphigus (PNP) and follicular dendritic cell sarcoma (FDCS) are closely associated with CD. It has been reported that 10-32% of patients with CD develop PNP (6-8), while 7-10% of FDCS cases are associated with hyaline-vascular CD (9). However, the coexistence of CD with both PNP and FDCS remains exceedingly uncommon. Herein, we describe a rare case of pelvic unicentric CD complicated by concurrent PNP and low- to intermediate-grade FDCS, emphasizing the diagnostic challenges and clinical implications of this unique association.

## Case report

A 67-year-old woman was admitted to HuiYa Hospital of The First Affiliated Hospital of Sun Yat-sen University (Huizhou, China) in July 2024 with an oral ulcer and systemic purple skin spots that had persisted for 1 month and a pelvic mass that had persisted for 5 days. The patient had presented with bilateral conjunctival hyperemia 1 month earlier, which gradually progressed to systemic purple skin spots and oral ulcers. The condition of the patient worsened: Ulcers developed on the lips and external genitalia, prompting the patient to seek medical attention. The patient had a 10-year history of type 2 diabetes. The latest hypoglycemic regimen is: miglitol (50 mg once daily) and liraglutide. The starting date for liraglutide treatment is July 2024 and the treatment dose is 0.6 mg once daily, and it will be gradually increased to 1.2 mg once daily. Marriage and childbearing history were unremarkable and there were no patients with similar diseases in the family.

*Physical examination (July 2024).* A general physical examination on admission revealed no abnormalities plus a pulse

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oxygen saturation of 98% on room air. Palpation revealed no superficial lymphadenopathy. Cardiovascular, pulmonary and abdominal examination results were normal. A dermatological evaluation revealed that the skin lesions were predominantly located in the mucosal areas. The oral mucosa exhibited erosions with white exudates, while the perioral region exhibited erosions, ulcerations and crusting with a noticeable mouth-opening limitation (Fig. 1A). An examination of the external genitalia revealed erosive and ulcerative lesions (Fig. 1B). Furthermore, multiple purple skin spots with variable morphologies and well-defined borders were noted throughout the body (Fig. 1C).

**Laboratory and imaging findings.** Laboratory findings (July 2024) were as follows: Blood glucose, 10.49 mmol/l (normal range, 3.9-6.1 mmol/l); fibrinogen, 4.05 g/l (normal range, 2.0-4.0 g/l); serum albumin, 36.0 g/l (normal range, 35-50 g/l); neutrophils,  $6.65 \times 10^9/l$  (normal range,  $1.8-6.3 \times 10^9/l$ ); normal white blood cells (normal range,  $3.5-9.5 \times 10^9/l$ ), red blood cells (normal range,  $3.8-5.1 \times 10^9/l$ ); platelet counts (normal range,  $125-350 \times 10^9/l$ ); hemoglobin, 137 g/l (normal range, 115-150 g/l); and C-reactive protein, 25.96 mg/l (normal range, 0.0-10.0 mg/l). The erythrocyte sedimentation rate was normal (normal range, 0-20 mm/h). The creatinine level (normal range, 53-115  $\mu\text{mol/l}$ ) and estimated glomerular filtration rate were normal (normal range, 90-120 ml/min/1.73 m<sup>2</sup>) and the urine protein levels were negative. The human chorionic gonadotropin (normal range, 0-6.00 mIU/ml) and estradiol levels were normal (normal range,  $\leq 138$  pg/ml). Human papillomavirus analysis (July 2024; PCR) was positive for the high-risk PV56 genotype. HIV, hepatitis virus antibodies and syphilis-specific antibodies were negative. The tumor marker levels were within normal limits [carcinoembryonic antigen,  $< 5.00$  ng/ml;  $\alpha$ -fetoprotein,  $< 20.00$  ng/ml; carbohydrate antigen (CA)15-3,  $< 35.00$  KU/l; CA199, 0-35.00 U/ml; CA125, 0-35.00 U/ml; CA72-4, 0-6.9 U/ml; cytokeratin 19 fragment, 0-3.30 ng/ml; neuron-specific enolase, 0-13.00 ng/ml]. Screening for pemphigus-related autoantibodies, including anti-bullous pemphigoid antigen (BP)180, BP230, desmoglein (Dsg)1 and Dsg3, exhibited anti-desmoplakin 3, anti-desmoplakin 1 and anti-BP230 antibody positivity. Antinuclear antibodies and specific autoantibodies (ANA, anti-double-stranded DNA antibody, anti-nRNP/Sm, anti-Sm, anti-SSA, anti-Ro-52, anti-SSB, anti-Scl-70, anti-JO-1) associated with connective tissue diseases were absent. IL-10 levels were 27.81 pg/ml (normal range, 0-10 pg/ml) and parathyroid hormone levels were 87.41 pg/ml (normal range, 15-60 pg/ml), while normal levels were obtained for IL-6 (normal range, 0-7 pg/ml), C3 (normal range, 0.5-2.8 g/l)/C4 (normal range, 0.1-0.4 g/l)/IgM (normal range, 0.3-2.2 g/l)/IgG (normal range, 8.6-17.4 g/l)/IgA (normal range, 1-4.2 g/l) and anti-Müllerian hormone (normal range, 0-0.39 ng/ml).

Laryngoscopy (July 2024) revealed erosions and white pseudomembranous exudates (Fig. 2A). Chest CT (July 2024) revealed bronchitis and bilateral mild bronchial dilation (Fig. 2B). No enlarged mediastinal lymph nodes were observed. Pelvic CT (July 2024) (plain and contrast-enhanced) revealed a solitary mass measuring  $\sim 114 \times 96 \times 116$  mm in the left pelvic region. The lesion had iso- to low-density characteristics with heterogeneous enhancement and contained multiple

patchy and non-enhancing necrotic areas (Fig. 2C). The venous drainage of the lesion converged into the left ovarian vein without enlarged lymph nodes. The liver and spleen were normal in size. Transvaginal ultrasonography (July 2024) revealed a hypoechoic mass measuring  $\sim 114 \times 86 \times 118$  mm in the anterosuperior aspect of the left side of the uterus (Fig. 2D).

**Pathology and immunohistochemistry (pelvic mass).** The findings of a histopathological examination of the pelvic mass were consistent with CD with focal progression to low- to intermediate-grade FDACS. H&E staining revealed the left side exhibited a concentric ('onion-skin') configuration with attenuation of lymphoid follicles, whereas the right side revealed effacement of the normal follicular architecture with replacement by a compressive mass consistent with progression to an FDC tumor (Fig. 3A). Immunohistochemical staining revealed the following (August 2024): Positive (focal or scattered) for CD21, CD23, CD35, C-X-C motif chemokine ligand 13 (data not shown) and CD68 (scattered; data not shown); negative for podoplanin, somatostatin receptor 2, CD20, CD3, CD1a, anaplastic lymphoma kinase, CD30, S-100, smooth muscle actin, CD117, melanin A, human melanoma black 45, CD34, cytokeratin, desmin and Epstein-Barr virus-encoded RNAs; and a Ki-67 proliferation index of  $\sim 10\%$  (data not shown). The results described in this paper are based on the pathological reports. Some immunohistochemical results are non-accessible image files, such as CXCL13, CD68 and Ki-67, etc. The immunohistochemical results suggested the focal progression to low- to intermediate-grade FDACS (Fig. 3B-D). The specific experimental materials and procedures can be found in the supplementary methods and Tables SI and SII.

**Diagnosis.** The patient was diagnosed in August 2024 with unicentric CD (UCD; hyaline-vascular) with PNP and FDACS (7). CT evaluation of the patient revealed a solitary large mass within the pelvic region, with no evidence of additional masses or lymphadenopathy elsewhere. Histopathological examination was consistent with CD, fulfilling diagnostic criteria for involvement confined to a single lymph node station. Based on H&E staining features, the lesion was classified as the hyaline-vascular variant. In the diagnostic workup for PNP, infectious, metabolic, and primary neurological disorders were initially excluded. The detection of pemphigus-associated autoantibodies served as a key diagnostic indicator, and in conjunction with the identified tumor, confirmed the diagnosis. The diagnosis of FDACS was established primarily based on histopathological characteristics observed on H&E staining, supported by positive immunohistochemical staining for CD21, CD25 and CD35.

**Treatment.** After admission, the patient was treated with intravenous methylprednisolone succinate (40 mg/dose once daily, intravenous drip), Human immunoglobulin (pH4) [20 g/dose once daily for a 4-day treatment course, intravenous drip (July 2024)] and thalidomide tablets (50 mg/evening, orally administered). Following the aforementioned treatments, a slight improvement was noted in the mucosal ulceration with reduced trismus severity. In July 2024, the patient underwent an exploratory laparotomy, pelvic tumor resection, adhesiolysis of the abdominal cavity and pelvic adhesiolysis. Postoperatively,



Figure 1. The skin symptoms of the patient. (A) Lips of the patient were ulcerated, with black crust formation and yellow secretion exudation. (B) White scurf on the labia majora and ulceration on the labia minora side. (C) Angular purplish purple skin spots with a clear boundary is seen in perineum area. (D) The appearance of the improved lip after treatment (from July to August 2024).

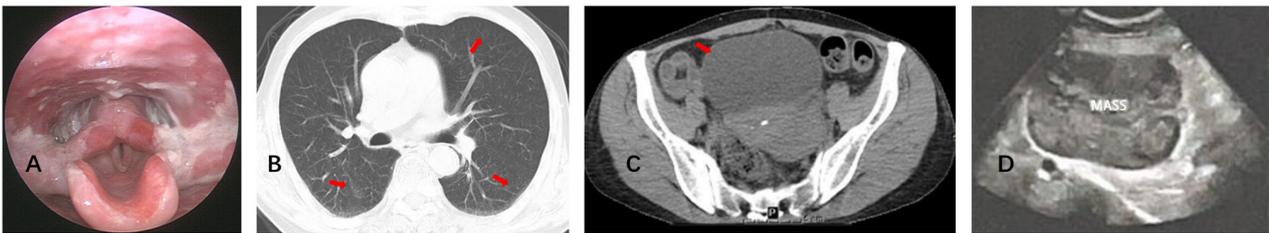


Figure 2. Laryngoscopy and imaging findings. (A) Erosions and white pseudomembranous exudates on laryngoscopy. (B) The imaging revealed bronchitis and mild bronchial dilation in both lungs (indicated by red arrows). (C) Pelvic CT plain scan exhibited a solitary mass in the left pelvic region (indicated by red arrow). (D) Transvaginal ultrasonography showing a hypochoic mass measuring ~114x86x118 mm.

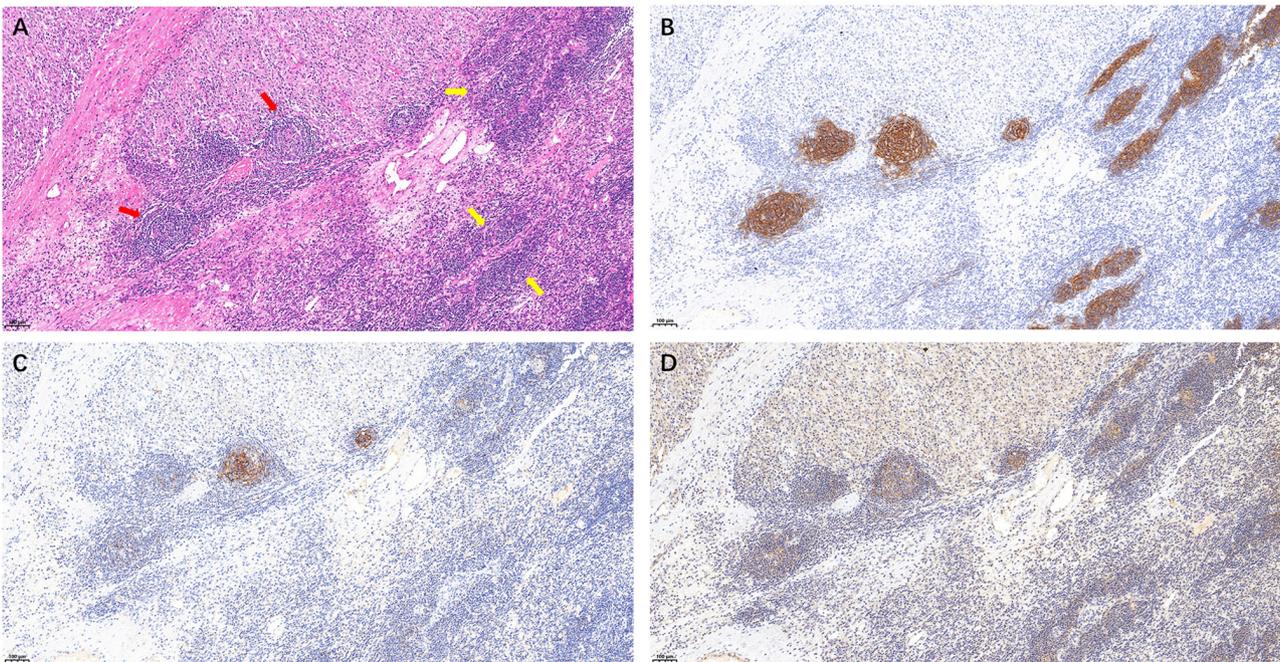


Figure 3. Pathological manifestations of tumors. (A) H&E staining demonstrated regional differences: The left side exhibited a concentric ('onion-skin', which marked with red arrows) configuration with attenuation of lymphoid follicles, whereas the right side exhibited effacement of the normal follicular architecture with replacement by a compressive mass consistent with progression to a follicular dendritic cell tumor (which marked with yellow arrows). (magnification, x100; scale bar, 100  $\mu$ m). Immunohistochemical staining of the pelvic mass demonstrated dark areas of (B) CD21, (C) CD23 and (D) CD35, which were focally positive, suggesting follicular dendritic cell sarcoma (magnification, x100; scale bar, 100  $\mu$ m).

the patient received antibiotic therapy (cefuroxime sodium for injection 1.5 g, twice a day, lasting for 2 days) and continued corticosteroid treatment (intravenous methylprednisolone succinate, 40 mg/dose once daily, lasting for 18 days) along

with supportive care. A tacrolimus ointment (0.1%, twice a day) was applied to the lips. Postoperative complications included hypoalbuminemia (albumin level, 22.4 g/l), which was managed with an intravenous human albumin infusion

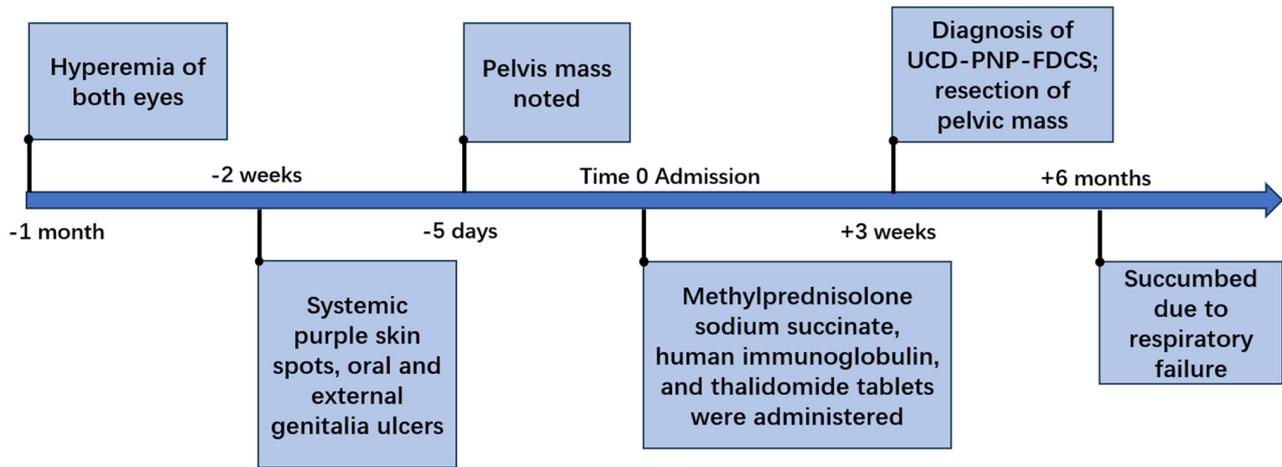


Figure 4. Timeline of the clinical course of the patient. The minus sign represents the time before admission and the plus sign represents the time after admission. PNP, paraneoplastic pemphigus; UCD, unicentric Castleman disease; FDSCS, follicular dendritic cell sarcoma.

(10 g/dose twice daily for 6 days). Upon the confirmation of CD through a pathological examination of the pelvic tumor, the treatment was as follows: Thalidomide 100 mg orally once nightly, cyclophosphamide 300 mg/m<sup>2</sup> orally once a week and prednisone 1 mg/kg orally twice a week.

**Outcomes.** After treatment (August 13th, 2024), the neutrophil count of the patient ( $5.12 \times 10^9/l$ ), and C-reactive protein (9.15 mg/l) and albumin levels (39.6 g/l) normalized. The generalized purple skin spots markedly subsided compared with that at admission and the oral and vulvar ulcers improved, although the erosions and pain persisted (Fig. 1D). The surgical incision healed well with normotension, reduced facial edema and decreased lip crusting. Despite the initial postoperative improvements, subsequent telephone follow-up (January 20th, 2025) revealed that the patient had ultimately succumbed due to respiratory failure. The timeline of the condition of the patient is shown in Fig. 4.

## Discussion

UCD typically involves a single lymph node and often presents without systemic symptoms. UCD most commonly occurs in the mediastinum, accounting for ~29% of all cases, followed by single lymph nodes or nodal regions in the cervical, abdominal and retroperitoneal areas (10). UCD also arises in rare sites such as the adrenal gland, liver, paravertebral region and breast (11-14). UCD is generally asymptomatic; however, when lesions are large or located in sensitive regions, patients may develop mass-effect symptoms such as dysphagia or localized pain and a minority may exhibit systemic manifestations including fever, night sweats, weight loss or anemia (15). MCD is characterized by lymphadenopathy in multiple regions and is further subdivided into HHV-8<sup>+</sup> and HHV-8<sup>-</sup> (idiopathic) types. Unlike those with UCD, patients with MCD exhibit lymphadenopathy and commonly present with systemic manifestations such as fever, night sweats, fatigue, weight loss, anemia, hepatic dysfunction, renal impairment and volume overload (for example, generalized edema, pleural effusion and ascites) (16). Both UCD and MCD can be complicated by

PNP and bronchiolitis obliterans (BO), which are associated with a poor prognosis (7,10,17).

PNP is a severe autoimmune blistering disease that primarily involves the oral mucosa and skin, and often initially presents as persistent oral ulcers or erosions. Other mucosal sites, such as the conjunctiva and nasal passages, may also be affected along with the skin, where polymorphic bullae and erythema are common (18). The respiratory system is frequently involved, with ~54% of patients exhibiting respiratory symptoms, including pulmonary dysfunction and, in certain cases, respiratory failure (18). FDSCS is a rare low- to intermediate-grade sarcoma that typically presents as a slow-growing, painless and localized mass (9). Most patients are asymptomatic and local symptoms usually arise only when the tumor compresses the adjacent structures. Respiratory system involvement is rare. On H&E staining, the tumor cells of FDSCS displayed abundant pale to eosinophilic cytoplasm, oval to elongated spindle-shaped nuclei, occasional cytoplasmic vacuolation and small nucleoli. Immunohistochemically, FDSCS typically expresses follicular dendritic cell markers including CD21, CD23 and CD35; the diagnosis is established by associating these characteristic morphologic features with a supportive immunophenotype (9,19,20).

The present study further reviewed case reports and meta-analyses of UCD coexisting with FDSCS and PNP. These findings suggested that adults with UCD as well as FDSCS or PNP generally do not present with notable laboratory abnormalities and that CD complicated by PNP may not exhibit positive pemphigus-related autoantibodies. Due to the complexity and difficulty of diagnosing CD, international evidence-based consensus diagnostic and treatment guidelines were promulgated in previous years (7,10,16). The diagnosis of CD and its associated complications depends primarily on a biopsy and histopathological examination (3,21). Furthermore, these guidelines advocate comprehensive laboratory and imaging evaluations to exclude conditions that may produce CD-like changes such as infections, lymphomas and polyneuropathy, organomegaly, endocrinopathy, monoclonal protein and skin changes syndrome (10).

Surgical resection, the treatment of choice for UCD, is often curative. Surgical intervention not only enables complete excision of the CD lesion, but it also mitigates the associated hyperinflammatory state (10,22). In patients with UCD and marked inflammation whose lesions are initially unresectable, treatment regimens extrapolated from iMCD, such as siltuximab in combination with glucocorticoids or the thalidomide-cyclophosphamide-prednisone protocol, may be employed (22). Surgical excision should be performed if medical therapy induces sufficient lesion regression for complete removal (10). However, patients with UCD and concomitant PNP and BO tend to have a worse prognosis. In cases of UCD associated with PNP, international consensus and guidelines recommend surgical resection (7,10). Nonetheless, even when the primary lesion is controlled, PNP frequently leads to respiratory complications (for example, BO), which may result in fatal respiratory failure in certain patients (7,23). Timely intervention is necessary and lung transplantation is an option. Complete surgical resection is the preferred treatment option for patients with FDCS. In cases that cannot be cured or show recurrence, adjuvant radiotherapy, chemotherapy and targeted immunotherapy may be considered; however, large-scale evidence to support these treatment methods is currently lacking (9).

Due to the rarity of CD coexisting with FDCS, survival outcomes are referenced from FDCS cases, which report a median progression-free survival of ~21 months and median overall survival (OS) of ~50 months (24). CD with concomitant PNP is associated with a poor prognosis; patients with PNP have 1- and 3-year OS rates of 76.9 and 57.6%, respectively, compared with 98.6 and 88.3% in those without PNP ( $P < 0.001$ ) (8). The development of BO is the key adverse prognostic factor in PNP (8,25).

According to the Consensus Statements of Deployment-Related Respiratory Disease and Chinese Guidelines for Paraneoplastic Pemphigus, patients with CD should undergo regular pulmonary function testing (PFTs). A decline in diffusing capacity warrants further evaluation using high-resolution chest CT and, if indicated, a lung biopsy to confirm or exclude BO (26,27). The European Guidelines further advise complete PFTs (including diffusing capacity of the lungs for carbon monoxide), inspiratory-expiratory high-resolution CT, 6-min pulmonary walk test or cardiopulmonary exercise examination (when available) and arterial blood gas analysis at diagnosis (27). For patients with active disease or progressive respiratory symptoms, symptom review and spirometry every 3 months are recommended, with repeat high-resolution CT or additional examinations (for example, cardiopulmonary exercise examination or surgical biopsy) if a rapid forced expiratory volume in 1 sec decline or worsening symptoms occur. In clinically stable patients, PFT intervals may be extended to every 6 months; in long-term stable cases, it can be extended to 6-12 months (28). These observations underscore the importance of guideline-directed dynamic surveillance for BO in patients with confirmed CD and/or PNP.

The present case represents a rare instance of CD associated with PNP and FDCS. Upon admission, the patient was treated with methylprednisolone, thalidomide and immunoglobulin to alleviate the inflammation along with

mucosal care of the lips and perineum. Surgical resection was performed after the oral mucosal condition of the patient was deemed tolerable for intubation. Upon the confirmation of CD through a pathological examination of the pelvic tumor, the treatment was as follows: Thalidomide 100 mg orally once nightly, cyclophosphamide 300 mg/m<sup>2</sup> orally once a week and prednisone 1 mg/kg orally twice a week. However, during subsequent follow-up, the patient succumbed due to respiratory failure and obstructive bronchiolitis was included in the mortality diagnosis as reported by the family.

In conclusion, CD is a key cause of PNP and BO. Complete surgical resection of the enlarged lymph nodes is the key treatment for patients with UCD and PNP. Therefore, monitoring for postoperative BO is necessary. If a patient is further diagnosed with BO, a lung transplantation can restore lung function and improve the prognosis. Several previous studies have demonstrated that patients experienced pulmonary function normalizing following surgery, with nearly complete resolution of oral mucosal lesions and marked improvement in health-related quality of life (21,29). In addition, patients who underwent a lung transplantation achieved a notable survival benefit compared with non-transplant with a 5-year survival rate of ~62.5% and no recurrence of CD was observed after transplantation. Although postoperative infection remained the predominant complication, the overall findings support lung transplantation as an effective therapeutic option for patients with end-stage disease (30,31).

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#### Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

#### Authors' contributions

YL and FZ obtained and analyzed the information of the patient and wrote the manuscript. ZW, GZ and QZ obtained and analyzed the information of the patient and reviewed the discussion part of the clinical diagnosis and treatment. QZ partially revised the article and generated the figures, and confirmed the authenticity of all the raw data. YL and FZ confirm the authenticity of all the raw data. All authors read and approved the final manuscript.

#### Ethics approval and consent to participate

The present case report was approved by The HuiYa Hospital of The First Affiliated Hospital, Sun Yat-sen University (Huizhou, China; approval no. 2025-0519-001).

## Patient consent for publication

Written informed consent to publish the present case information and accompanying images was obtained from the patient and their family.

## Competing interests

The authors declare that they have no competing interests.

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