

Case Report

Primary Small Bowel MALT Lymphoma with Concurrent Pulmonary Dissemination: A Case Report

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Abstract

Marginal zone lymphomas (MZLs) are indolent mature B-cell neoplasms. Approximately 3.4% of gastrointestinal mucosa-associated lymphoid tissue (MALT) lymphomas involve the small bowel. Pseudoaneurysmal dilation has been reported in up to 35% of patients with small bowel MALT lymphoma. We report the case of a 73-year-old woman with ulcerative colitis in remission who presented with hematuria, constitutional symptoms, and progressive respiratory distress. Imaging incidentally revealed pseudoaneurysmal dilation of the small bowel and thoracic findings suggestive of pulmonary lymphangitic dissemination. A PET scan showed lung, nodal, and small bowel infiltration. Histopathological and flow cytometry examinations confirmed small bowel MZL, and bone marrow biopsy excluded marrow involvement. The patient was treated with R-CHOP chemotherapy and then R-Bendamustine, achieving complete clinical and radiological remission. This case illustrates a rare presentation of intestinal MALT lymphoma and emphasizes the diagnostic significance of correlating imaging and clinical findings in identifying pseudoaneurysmal dilatation and distinguishing it from other potential causes.

Keywords: MALT lymphoma; marginal zone B-cell lymphoma; pseudoaneurysmal dilation

1. Introduction

Marginal zone lymphomas (MZLs) are indolent mature B-cell neoplasms, representing 7% of non-Hodgkin lymphomas. The main subtypes include: splenic (SMZL, 9%), nodal (NMZL, 30%), and extranodal marginal zone lymphoma of mucosa-associated lymphoid tissue (MALT), which represents the most frequent subtype (61%), as well as primary cutaneous, and pediatric forms [1,2].

MALT lymphoma are strongly associated with chronic antigenic stimulation related to infectious diseases (e.g., *Helicobacter pylori* in gastric MALT, *Campylobacter jejuni* in the small intestine, Epstein–Barr virus) and/or autoimmune diseases (e.g., ulcerative colitis, Crohn’s disease, Sjögren’s syndrome or IgG4-related disease). The chronic inflammatory environment increases the risk of chromosomal abnormalities, monoclonal or oligoclonal B-cell expansion, and ultimately lymphoproliferative disorders [3].

In a large observational study conducted by Khalil et al. in 2014, including 13,096 patients, 67.3% were diagnosed with extranodal MALT lymphoma [4]. Among these cases (*n* = 8821), the most frequently involved sites were the stomach (30.6%), spleen (12.7%), eye and ocular adnexa (11.6%), skin (7.8%), lungs (7.7%), salivary glands (7.2%), and colon and



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rectum (4.5%). Small intestinal MALT lymphoma accounted for 2.9% of all extranodal marginal zone lymphomas ($n = 258$) [4]. Small bowel involvement is uncommon, and the symptoms include abdominal pain, bloating, and malabsorptive diarrhea. The most common image finding is aneurysmal dilatation with associated bowel wall thickening secondary to lymphomatous infiltration [5,6].

Given its high prevalence and strong association with *Helicobacter pylori* infection, eradication therapy using triple or quadruple regimens constitutes the first-line treatment for gastric MALT lymphoma, achieving cure rates of up to 80% and complete remission within 12 months. Treatment protocols may vary according to local epidemiology and antimicrobial resistance patterns, and typically include a proton pump inhibitor combined with clarithromycin and amoxicillin or bismuth, metronidazole, and tetracycline for 10–14 days [7]. In contrast, *H. pylori* eradication therapy has not demonstrated efficacy in small intestinal MALT lymphoma and is therefore not currently recommended. Management of this entity usually relies on systemic chemo-immunotherapy, most commonly combining rituximab (a monoclonal anti-CD20 antibody) with alkylating agents such as cyclophosphamide or bendamustine, or with anthracycline-based regimens, achieving partial and complete remissions within 6–12 months. The role of radiotherapy in small intestinal MALT lymphoma is limited by bowel mobility, which compromises treatment precision, while surgical resection is generally reserved for complications including obstruction, perforation or severe dilatation [3].

Overall, MALT lymphoma is an indolent clinical course with a generally favorable prognosis; however, clinical outcomes may vary depending on the primary site, disease stage, and response to initial therapy [3].

Herein, we report a rare case of small bowel MZL, incidentally discovered during the study of hematuria and suspected due to the pseudo-aneurysmal dilation.

2. Case Presentation

A 73-year-old woman with a previous history of ulcerative colitis, treated with oral and rectal mesalazine and currently in clinical remission, presented to the emergency department with a self-limited episode of gross hematuria. Initial laboratory blood tests were unremarkable, and she was discharged with an outpatient referral to the urology department.

A subsequent contrast-enhanced computed tomography (CT) scan, performed in the ambulatory setting as part of the diagnostic workup for hematuria, revealed a pseudoaneurysmal dilatation involving the third and fourth portions of the duodenum and the proximal jejunum and left nephrolithiasis affecting the upper calyceal group. In addition, basal lung window images included in the abdominal CT demonstrated subtle peripheral ground-glass opacities in the lower lobes. The nephrolithiasis was ultimately identified as the cause of hematuria, while the intestinal findings and the incidental pulmonary abnormalities prompted further evaluation (Figure 1A,B). One week later, the patient came back to the emergency department after developing new-onset respiratory symptoms, including a productive cough with white sputum, asthenia, adynamia, and a previously unreported unintentional weight loss of approximately 6 kg over the preceding year.

The patient was admitted with a suspected diagnosis of small bowel lymphoma with possible pulmonary lymphangitic involvement, based on the attending physician's review of the previously obtained CT findings. A contrast-enhanced CT scan of the chest revealed multiple ground-glass infiltrates with thickening of the interlobular septa, predominantly in the peripheral regions of the lower and middle lobes, exhibiting an apicobasal gradient (Figure 1C,D). An initial positron emission tomography/computed tomography (PET/CT) showed gastrointestinal and pulmonary involvement with increased metabolic activity (SUVmax 9.31) and thickening in the wall of the third and fourth portions of the duodenum,

as well as hypermetabolic lymph nodes at the root of the mesentery and the splenic hilum (SUVmax 5.09). Additionally, bilateral pulmonary infiltrates, predominantly affecting the middle and lower lobes, exhibited low-grade metabolic uptake (SUVmax 3.47), which was considered radiologically suggestive of pulmonary involvement (Figure 1E).

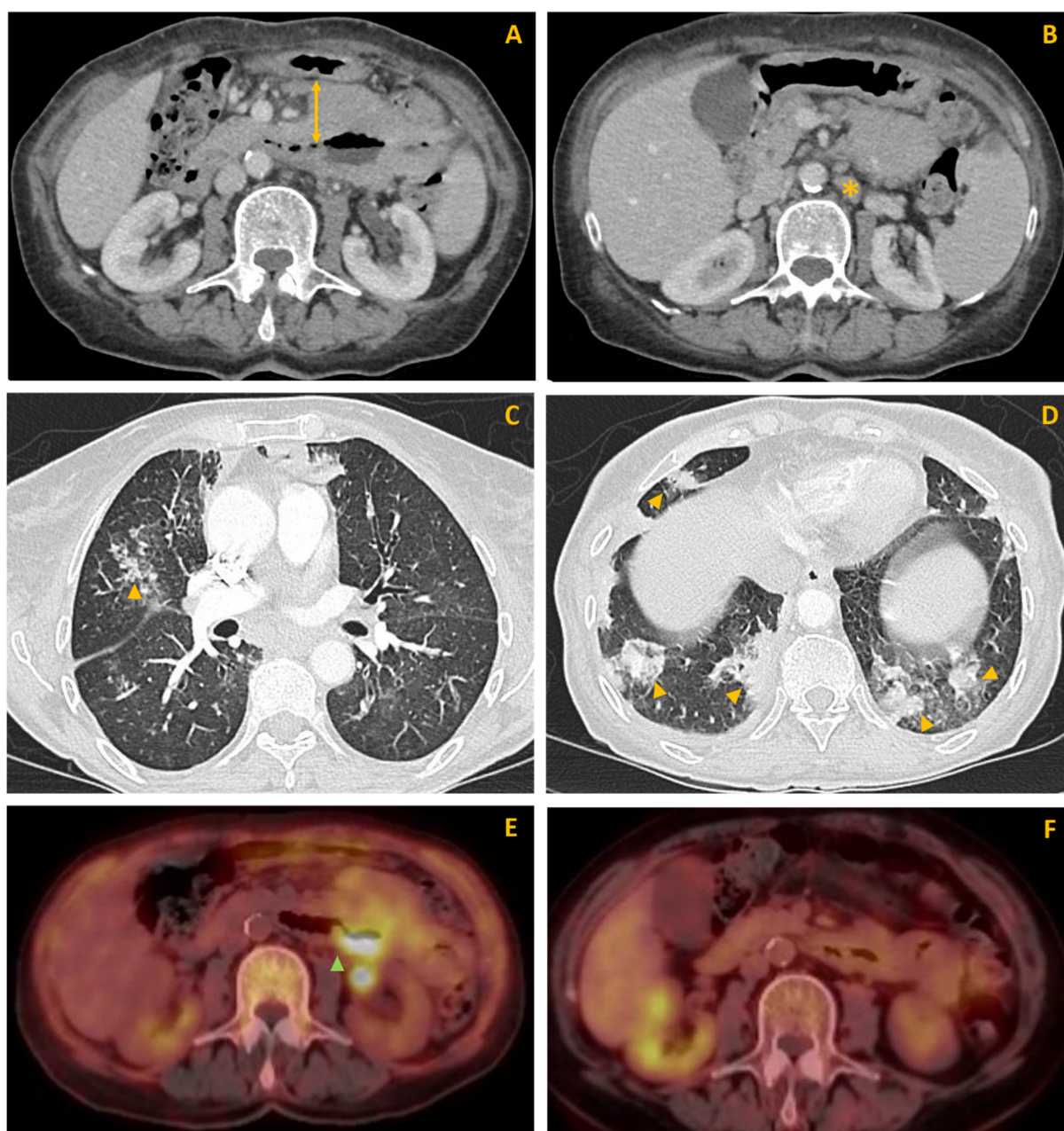


Figure 1. Image findings. (A) Abdominal CT shows circumferential thickening (20–35 mm; yellow arrow) of the third and fourth parts of the duodenum and proximal jejunum, with pseudoaneurysmal dilatation suggestive of lymphomatous involvement of the small bowel. (B) Lateral aortic lymph node enlargement (yellow asterisk). (C,D) Multiple alveolar opacities surrounded by ground-glass attenuation with thickened interlobular septa, affecting both lungs with subpleural distribution with a basal predominance. The findings are highly suggestive of pulmonary lymphomatous involvement (yellow arrowheads). (E) PET/TC shows mesenteric and retroperitoneal lymphadenopathy with duodenal thickening (green arrowhead). (F) First restaging PET/CT showed a partial metabolic response (Deauville 4) of the duodenal lesion and a complete response of the infradiaphragmatic lymph node.

Laboratory tests revealed a mild normocytic normochromic anemia (Hb 9.7 mg/dL), an elevated erythrocyte sedimentation rate adjusted by age (89 mm/h), and increased beta 2 microglobulin levels (3.5 µg/mL). LDH was normal (200 UI/L, normal range < 250 UI/L) (Table 1).

Table 1. Blood test results.

Test	Result	Unit	Reference Range
Hemoglobin	9.7	g/dL	12–15
Hematocrit	31.6	%	36–43
MCV	82.5	fL	86–100
MCHC	30.7	g/dL	31.5–34.5
RDW	14.8	%	11.2–15.2
Leukocytes	8.95	$\times 10^3/\mu\text{L}$	3.5–11.0
Neutrophils	5.7	$\times 10^3/\mu\text{L}$	1.5–8.8
Lymphocytes	2.5	$\times 10^3/\mu\text{L}$	1.0–5.0
Monocytes	0.7	$\times 10^3/\mu\text{L}$	0.1–1.0
Platelets	574	$\times 10^3/\mu\text{L}$	150–450
ESR	89	mm/h	Inf. 25
CRP	2.66	mg/dL	0.0–0.5
Albumin	2.8	g/dL	3.2–4.8
Calcium	8.0	mg/dL	8.8–10.2
Potassium	3.4	mmol/L	3.7–5.2
Phosphorus	3.2	mg/dL	2.4–5.1
Creatinine	0.5	mg/dL	0.5–0.9
GFR	95.4	mL/min/1.73 m ²	90–120
BUN	8	mg/dL	9–23
LDH	200	UI/L	Inf. 250
ALT	6	UI/L	10–49
AST	18	UI/L	0–34
Alkaline phosphatase	62	UI/L	45–129
Uric acid	3.8	mg/dL	2.4–5.7
Ferritin	146	ng/mL	-
TSAT	9	%	-
beta2 microglobulin	3.50	µg/mL	Inf. 3.00
AgS HBV	Negative	-	-
Ac HCV	Negative	-	-
HIV	Negative	-	-

MCV: mean corpuscular volume, MCHC: Mean Corpuscular Hemoglobin Concentration, RDW: Red Cell Distribution Width, TSAT: transferrin saturation index, ESR: erythrocyte sedimentation rate, CRP: C reactive protein, GFR: glomerular filtration rate, BUN: blood ureic nitrogen, LDH: lactate dehydrogenase, ALT: alanine aminotransferase, AST: aspartate aminotransferase, TSAT: transferrin saturation, AgS HBV: hepatitis b surface antibody, Ac HCV: hepatitis C antibodies, HIV: human immunodeficiency virus.

We discussed the patient with general surgery and oncology teams to determine the optimal strategy for obtaining a representative tissue sample. Initially, an upper gastrointestinal endoscopy was performed, and duodenal-jejunal biopsies were obtained, which were negative for malignancy. Endoscopic ultrasound was subsequently considered as an alternative diagnostic approach; however, diagnostic laparoscopy for lymph node sampling was deemed the most effective strategy, albeit technically more challenging.

During this period, the patient developed acute fever, cough, and dyspnea, requiring supplemental oxygen at 3 L/min to maintain oxygen saturation above 92%. Chest radiography and CT pulmonary angiography revealed a right lower pulmonary embolism, along with pulmonary consolidation and increased parenchymal nodular involve-

ment, suggestive of disease progression. Given this clinical scenario, bronchoscopy was not performed.

In this context, therapeutic anticoagulation with enoxaparin (60 mg every 12 h), broad-spectrum antibiotic therapy (piperacillin/tazobactam 4.5 g every 6 h), and high dose intravenous methylprednisolone pulse therapy (500 mg once daily for 5 days) were initiated following a careful risk–benefit assessment. This decision was based on the severity of acute pulmonary involvement, the rapid progression of inflammatory infiltrates, and the absence of overt infectious features or a definitive histological diagnosis.

Within the subsequent 72 h, the patient showed marked clinical improvement, with resolution of respiratory symptoms and discontinuation of oxygen therapy.

A laparoscopic lymph node biopsy confirmed the diagnosis of small bowel MZL with architectural distortion due to a prominent lymphocytic infiltrate composed primarily of B cells, immunoblasts, monocytes, and centrocytes within the marginal zone of reactive follicles, extending into the interfollicular regions. Atypical lymphoid cells tested positive for CD20, CD79 alpha, BCL2, PAX5, and CD43, while markers for CD3, CD4, CD5, Cyclin D1, CD10, and CD8 were negative (Figure 2). Lymph node immunophenotyping showed a CD19⁺/CD20⁺ B-cell population with CD5[−]/CD10[−]/CD23[−] phenotype and kappa light chain restriction, excluding small lymphocytic lymphoma. Prognostic indices were calculated, yielding 3 points for the International Prognostic Index (IPI) and 2 points for the MALT-IPI. During the staging process, bone marrow aspiration and biopsy were performed, both of which were negative for lymphomatous infiltration.

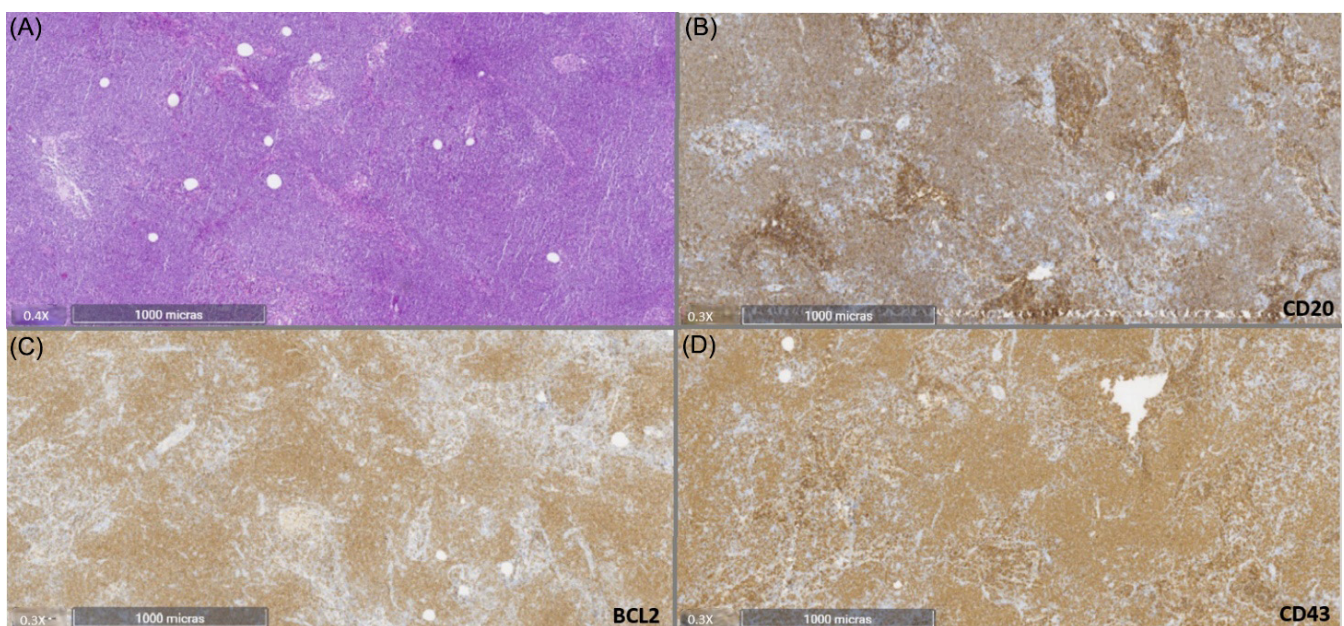


Figure 2. Retroperitoneal excisional lymph node biopsy. (A) Hematoxylin–eosin staining exhibits architectural distortion with a prominent lymphocytic infiltrate composed of B cells. Immunohistochemistry in atypical lymphoid cells is positive for CD20 (B), BCL2 (C), and CD43 (D). The pathologist found a heterogeneous population of dispersed immunoblasts, monocytes, and centrocytes located in the marginal zone of reactive follicles, extending into the interfollicular regions.

The patient was initially diagnosed at our institution. Given the advanced stage of disease (stage IV) and the aggressive clinical presentation, characterized by highly symptomatic pulmonary involvement, first-line treatment with the R-CHOP regimen (rituximab, cyclophosphamide, doxorubicin, vincristine, and prednisone) was selected (IPI score: 3; MALT-IPI score: 2). A baseline transthoracic echocardiogram was performed prior to treatment initiation due to planned anthracycline exposure.

Following transfer to her referral center and achievement of initial clinical remission (Figure 1F), the patient completed a total of six treatment cycles: one cycle of R-CHOP, followed by five cycles of bendamustine–rituximab (BR). Treatment response was monitored using PET/CT scans and assessed according to the Deauville criteria. The first restaging PET/CT, performed after the initial treatment phase (1 R-CHOP + 2 BR cycles), showed a Deauville score of 4, which was maintained on two subsequent PET/CT evaluations. The patient remains in complete remission 10 months after finishing therapy.

3. Discussion

MZL are rare primary gastrointestinal neoplasms. The patient presented herein showed an unusual clinical manifestation. She had a prior diagnosis of ulcerative colitis, a condition recognized as a potential risk factor for lymphoproliferative disorders. Patients with inflammatory bowel disease, including Crohn’s disease and ulcerative colitis, have an increased risk of lymphoma, which has been attributed to chronic immune stimulation and, in some cases, exposure to immunosuppressive therapies [8,9]. Although the available evidence remains controversial, a meta-analysis by Zamani et al. reported a 30% increased risk of lymphoma in patients with inflammatory bowel disease, irrespective of treatment exposure or lymphoma subtype (Hodgkin vs. non-Hodgkin lymphoma) [10].

This case highlights the relevance of imaging–clinical correlation in the diagnostic process. Cupps et al. first described the characteristic aneurysmal dilatation pattern in up to 35% of patients with small bowel lymphoma [11]. Key differential diagnoses include gastrointestinal stromal tumors, leiomyosarcoma, metastatic disease, and primary adenocarcinoma [12] (Table 2).

Multifocal disease was suspected based on clinical features, radiological findings on CT and PET/CT, and the patient’s favorable response to corticosteroid therapy. Given her acute respiratory deterioration, bronchoscopy was not performed, and pulmonary involvement could not be histologically confirmed. In published case series from Japan and the Netherlands, the reported prevalence of multifocal MALT lymphoma ranges from 2% to 32%, respectively [13,14]; however, concurrent pulmonary, nodal and small bowel involvement was not described in these cohorts. Lam et al. reported a rare case of multifocal gastric and pulmonary MALT lymphoma, confirmed by immunohistochemical analysis, underscoring the heterogeneity and diagnostic complexity of this entity [15].

In this clinical context, the optimal first-line therapeutic strategy remains uncertain, as evidence guiding treatment selection in advanced and aggressive presentations of MALT lymphoma is limited. While bendamustine–rituximab (BR) is generally preferred in more indolent or localized disease, the initial choice of R-CHOP was considered appropriate given the patient’s disease burden and severity of clinical presentation.

Table 2. Reported cases with small bowel pseudoaneurismal dilation.

Reference	Age	Sex	Symptoms	Imaging Findings	Biopsy
[16]	84	F	Hematochezia	Thickening of the small intestine	Small bowel stromal tumor
[17]	73	M	Intestinal obstruction, weight loss and abdominal pain	Pseudo-aneurysmal dilation of the last ileal loop, multiple coelio-mesenteric lymphadenopathy	Anaplastic large cell lymphoma type T

Table 2. Cont.

Reference	Age	Sex	Symptoms	Imaging Findings	Biopsy
[18]	23	M	Left axilar mass, weight loss and abdominal pain	Jejunum and ileum thickening, aneurismal dilatation, ileo-ileal jejunal invagination, and multiple lymphadenopathies	High-grade B cell lymphoma
[19]	43	F	Periumbilical abdominal pain, nausea and vomiting	Gastric wall thickening. Small bowel thickening and intussusception in the left iliac fossa	Gastric B-cell lymphoma
Actual case report	73	F	Productive cough, asthenia, adynamia, and unintentional weight loss	Pseudoaneurysmal dilatation of the third and fourth portions of the duodenum and proximal jejunum—pulmonary lymphomatous involvement	MALT

F: female, M: male.

4. Conclusions

Primary MALT lymphoma of the small bowel with concurrent lung involvement is exceptionally rare, especially when typical digestive symptoms are absent. This case illustrates the diagnostic challenges posed by atypical presentations, where characteristic imaging findings—such as pseudoaneurysmal dilatation—guided further investigation. The coexistence of ulcerative colitis highlights the potential role of chronic inflammatory conditions as predisposing factors. Comprehensive integration of clinical, radiological, and histopathological data was essential for accurate diagnosis, staging, and selection of an effective therapeutic strategy.

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Abbreviations

The following abbreviations are used in this manuscript:

MZL	Marginal zone lymphomas
MALT	Mucosa-associated lymphoid tissue
R-CHOP	Rituximab—Cyclophosphamide, Doxorubicin, Vincristine, Prednisone
EMZL	Extranodal Marginal zone lymphomas
CT	Computed tomography
PET/CT	Positron Emission Tomography/Computed Tomography

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