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【 CASE REPORT 】

Development of Eosinophilic Temporal Arteritis and Digital Ischemia in a Patient with Hypereosinophilic Syndrome

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Abstract:
We describe a case of eosinophilic temporal arteritis in a 61-year-old woman with hypereosinophilic syndrome, who developed subcutaneous nodules in the temporal areas and digital cyanosis with small nodules on the sides of her fingers. Ultrasound revealed occlusion and corkscrew-like changes of the temporal and digital arteries, respectively. Temporal artery biopsy revealed eosinophilic vasculitis without giant cell formation. Angiography showed occlusion of the ulnar and digital arteries. Administration of low-dose corticosteroid improved the temporal artery swelling and digital cyanosis. More reports of similar cases are required to characterize this type of non-giant cell eosinophilic vasculitis that affects the peripheral arteries.

Key words: eosinophilic temporal arteritis, digital ischemia, eosinophilia

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Introduction

Hypereosinophilic syndrome (HES) is a rare systemic disorder of unknown etiology characterized by persistent eosinophilia in blood (absolute eosinophil count >1,500/μL), which cannot be attributed to secondary causes of eosinophilia, as well as end-organ damage mediated by eosinophils, such as eosinophilic esophagitis, eosinophilic gastroenteritis, eosinophilic dermatitis, eosinophilic pneumonia, and eosinophilic myocarditis (1). In rare cases, HES can also involve the vascular systems, leading to eosinophilic vasculitis that affects not only the small vessels (e.g., cutaneous eosinophilic vasculitis) but also the medium-sized vessels of the limbs (2-11).

Giant cell arteritis (GCA) is large vessel vasculitis in the elderly (usually >50 years of age), which is pathologically characterized by mononuclear infiltration of the arterial wall and granuloma formation with multinucleated giant cells (12). Since GCA mostly occurs in the temporal arteries, it is also known as temporal arteritis. However, not all cases of "temporal arteritis" are GCA. Genereau et al. demonstrated that non-giant cell vasculitis, such as polyarteritis nodosa (PAN), eosinophilic granulomatosis with polyangiitis (EGPA), microscopic polyangiitis, granulomatosis with polyangiitis, cryoglobulinemic vasculitis, and rheumatoid vasculitis were identified in 4.5% of positive (inflamed) temporal artery biopsy specimens (13). Temporal artery involvement was also reported - albeit less frequently - in patients with juvenile temporal arteritis (JTA) (14), thromboangiitis obliterans (TAO, Buerger's disease) (15-19), HES (10), and other secondary vasculitis (20-28).

In this report, we describe a rare case of hypereosinophilia in an elderly woman who had corticosteroid-responsive temporal region swelling and digital ischemia due to non-granulomatous eosinophilic vasculitis. The simultaneous occurrence of eosinophilic temporal arteritis and digital ischemia is rare. One similar case has been reported in the literature by Ito et al. (10) and Motegi et al. (29), who considered that eosinophilic peripheral arteritis could be a new disease entity related to HES. Based on the case presentation and a review of the relevant literature, we discuss the possible differential diagnosis of peripheral eosinophilic arteritis.

Case Report

A 60-year-old Japanese woman was brought to our hospi-

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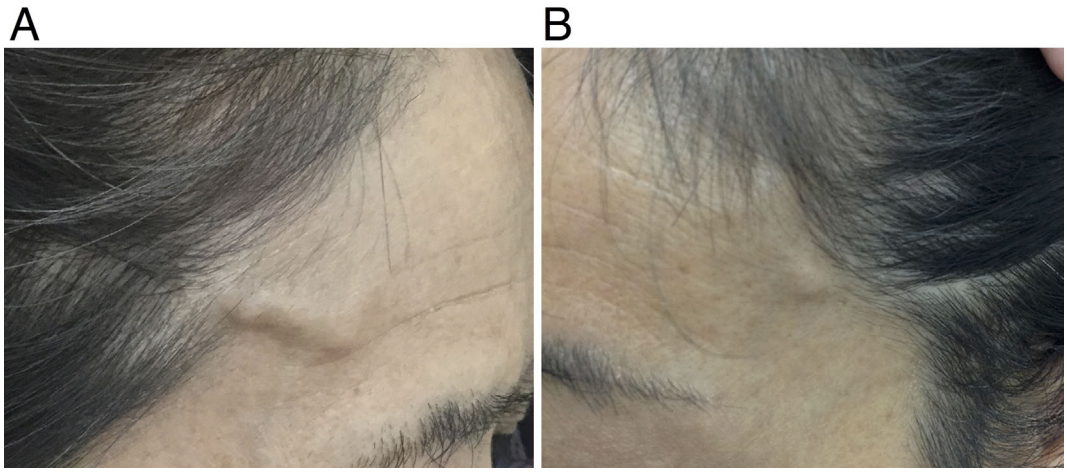

Figure 1. **Subcutaneous nodules in the temporal areas that were slightly pruritic and tender and considered to be swelling of the superficial temporal artery with reduced pulsation. A: The right temporal area. B: The left temporal area.**

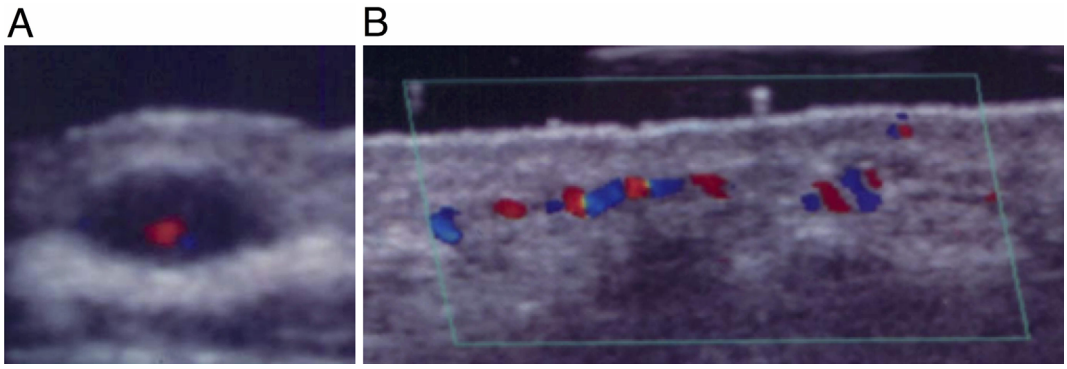

Figure 2. **Color-duplex ultrasound images of the affected vessels. A: Hypoechogenic wall thickening and luminal stenosis of the right temporal artery. B: The corkscrew architecture of the digital artery of the affected finger.**

tal in July with a one-month history of subcutaneous nodules in her temporal areas that were slightly pruritic and tender (Fig. 1). She had no other symptoms of note, in particular no systemic symptoms (fever, night sweats, and weight loss), visual impairment, or musculoskeletal symptoms suggestive of polymyalgia rheumatica, such as stiffness and pain in the neck, shoulder and hip areas. She was a smoker with a 20-30 pack-year smoking history. She had no history of dyslipidemia, diabetes mellitus, hypertension, or allergic diseases, including respiratory symptoms suggestive of bronchial asthma. She had a family history of neurofibromatosis, and her son had Café-au-lait lesions. Besides temporal artery enlargement, she reported having consistently noticed several subcutaneous nodules on her head.

A general physical examination revealed that her height was 148 cm, and her weight was 37 kg. Her body temperature was 35.7°C, her pulse rate was 61 beats per minute, and her blood pressure 131/71 mmHg. Her palpebral conjunctiva was not anemic, and her bulbar conjunctiva was not icteric. Cardiopulmonary and abdominal examinations revealed no abnormalities. No peripheral edema was noted. The subcutaneous nodule in her right temporal area, was considered to be the swelling of the superficial temporal artery with reduced pulsation, which was confirmed by ultrasound (US) imaging showing hypoechogenic wall thickening and luminal stenosis of the temporal artery (Fig. 2A). A complete blood count revealed a white blood cell count of 10,300 with 5.4% eosinophils (556/μL) (Table). Her erythrocyte sedimentation rate (ESR), and C-reactive protein (CRP), complement 3 (C3), complement 4 (C4), immunoglobulin G, immunoglobulin E, fibrin degradation product (FDP), and D-dimer levels were within the normal ranges. A microscopic stool examination was negative for ova and parasites. Her antinuclear antibody (Ab) titer was 1:40 (normal value, <1:40). Additionally, tests for rheumatoid factor, anti-phospholipid Abs, anti-deoxyribonucleic acid Abs, anti-myeloperoxidase (MPO) Abs, and anti-proteinase-3 Abs were negative. The patient's serum was negative for hepatitis B and C including hepatitis B surface antigen (HBsAg), hepatitis B surface antibody (anti-HBs), hepatitis B core antibody (anti-HBc), and anti-hepatitis C virus antibody (anti-HCV). Ischemic lesions suggestive of GCA, such as arteritic

Table. The Laboratory Values on Admission for the Patient with Eosinophilic Arteritis.

Laboratory tests	Results	Reference range
CBC		
WBC count	8,300	2,700-8,800 /μL
Eosinophil (%)	15.0	0-7 %
RBC count	458	370-540 $\times 10^4$/μL
Hemoglobin	14.5	11.0-17.0 g/dL
Hematocrit	42.2	34.0-49.0 %
Platelets	27.5	14-34 $\times 10^4$/μL
Coagulation-fibrinolysis		
PT-INR	0.94	0.85-1.15
APTT	35.5	30.0±5.0 s
Fibrinogen	324	200-400 mg/dL
D-dimer	0.15	<0.8 μg/mL
FDP	14	<2.8 μg/mL
Biochemistry		
Total protein	6.9	6.6-8.2 g/dL
Albumin	4.4	3.9-4.9 g/dL
AST	19	8-38 U/L
ALT	26	4-44 U/L
LDH	153	106-211 U/L
γ-GTP	28	17-73 U/L
Alkaline phosphatase	209	104-338 U/L
Total bilirubin	0.45	0.2-1.2 mg/dL
BUN	8.1	8.0-22.6 mg/dL
Serum creatinine	0.54	0.4-0.8 mg/dL
CK	69	43-165 U/L
ESR	12.0	<15 mm per hour
CRP	<0.3	<0.3 mg/dL
Urinalysis		
Proteinuria	(-)	(-)
WBC	<1/HPF	<1-4/HPF
RBC	<1/HPF	<1-4/HPF
Casts	(-)	(-)
Immunological		
IgG	1,041	870-1,700 mg/dL
IgA	127	110-410 mg/dL
IgM	169	35-220 mg/dL
IgE	82.8	<270 IU/mL
C3	96	65-135 mg/dL
C4	21	13-35 mg/dL
ANA	1:40 (homogeneous, speckled)	<1:40
RF	6.0	<15 U/mL

CBC: complete blood count, WBC: white blood cell, RBC: red blood cell, PT-INR: prothrombin time-international normalized ratio, APTT: activated partial thromboplastin time, s: seconds, FDP: fibrin degradation product, AST: aspartate aminotransferase, ALT: alanine aminotransferase, LDH: lactate dehydrogenase, γ-GTP: γ-glutamyl transpeptidase, BUN: blood urea nitrogen, CK: creatine kinase, ESR: erythrocyte sedimentation rate, CRP: C-reactive protein, HPF: high power field, IgG: immunoglobulin G, IgM: immunoglobulin M, IgA: immunoglobulin A, IgE: immunoglobulin E, C3: complement 3, C4: complement 4, ANA: anti-nuclear antibody, Ab: antibody, RF: rheumatoid factor

ischemic optic neuropathy and central retinal artery occlusion (30), were absent on an ophthalmological examination. Since her ESR and CRP levels were not elevated, the patient was followed-up without medication.

During follow-up, she complained of worsening pain of the swollen temporal arteries. Although her ESR, CRP, D-dimer, and FDP levels remained within the normal ranges, the absolute eosinophil count (AEC) gradually increased to 742/μL in August, 850/μL in September, and 1,954/μL in October, when she began to experience coolness of the fin-

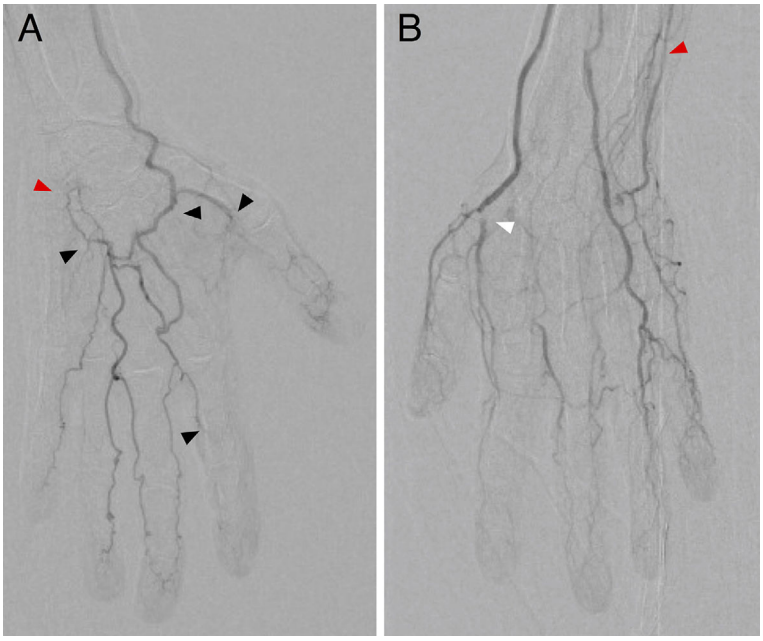

Figure 3. **Angiography showing the occlusion of the ulnar artery and digital arteries. A: The right hand. The occluded portions of the ulnar and palmar digital arteries are indicated by red and black arrows, respectively. B: The left hand. The occluded portion of the palmar arch artery and narrowing of the ulnar artery are indicated by white and red arrows, respectively. Decreased blood flow in the digital arteries is also noted.**

gers of both hands accompanied by cyanotic discoloration of her right ring and left index fingers. Computed tomography (CT) of the chest and echocardiography revealed no abnormalities. She also noticed small and slightly pruritic subcutaneous nodules around the lateral sides of her fingers. Color duplex ultrasonography of the hands showed occlusion of the ulnar arteries, and corkscrew architecture of all the digital arteries except the right index finger (Fig. 2B). Angiography showed occlusion of the ulnar artery, palmar arch artery, and proper palmar digital arteries (Fig. 3). Temporal artery biopsy of the right superficial temporal artery showed intense transmural infiltration of inflammatory cells, mainly composed of eosinophils, which involved all three layers of the arterial wall (intima, media, and adventitia; Fig. 4D-F), intimal hyperplasia (Fig. 4A), and disruption of the internal elastic lamina (Fig. 4C). However, there was no giant cell formation. Capillary proliferation was also noted, suggesting repeated occlusion and recanalization of the artery.

The administration of prednisolone (20 mg/day) improved the acrocyanosis of both hands with the normalization of the AEC. Thus, the AEC rapidly decreased to 25/μL after two weeks of corticosteroid treatment. Three months later when the prednisolone dose was reduced to 15 mg per day, the swelling of the remaining temporal arteries decreased. Although there were several small subcutaneous nodules in the occipital area, the biopsy specimens showed spindle-shaped cells proliferating in waves with mucoid substance accumulation, embedded within a fibrillary collagenous matrix, which was compatible with neurofibroma. Eventually, the prednisolone dose was reduced to 1 mg per day over 4 years. Meanwhile, there was no relapse of temporal arteritis, occurrence of ophthalmological problems or appearance of blood hypereosinophilia. We did not perform a second angiographic evaluation because the patient did not consent to it.

Discussion

We described the case of non-granulomatous eosinophilic vasculitis of the temporal arteries in an elderly woman with peripheral blood eosinophilia complicated by digital ischemia due to ulnar artery occlusion at the wrist-level. We considered that the patient in the present case had eosinophilic vasculitis, based on the transluminal infiltration of eosinophils with disruption of the elastic lamina. Previous studies revealed that PAN, which is characterized by necrotizing inflammation-usually involving middle-sized arteries-can affect the temporal arteries (31, 32). Of note, Oiwa et al. reported an intriguing case with HES secondary to PAN, in which the patient developed marked blood hypereosinophilia and eosinophil-rich medium-sized vasculitis (33). However, massive infiltration of eosinophils in the vessel walls is typically considered to be an unusual finding in PAN (11). In addition, the structure of the smooth muscle in the media was maintained in the present case, even though the elastic lamina was disrupted. Thus, the possibility of PAN was considered to be low in our case. Considering that the main pathological finding of the present case was eosinophilic vasculitis affecting the temporal arteries, the differential diagnosis could include JTA with eosinophilia (JTAE), EGPA,

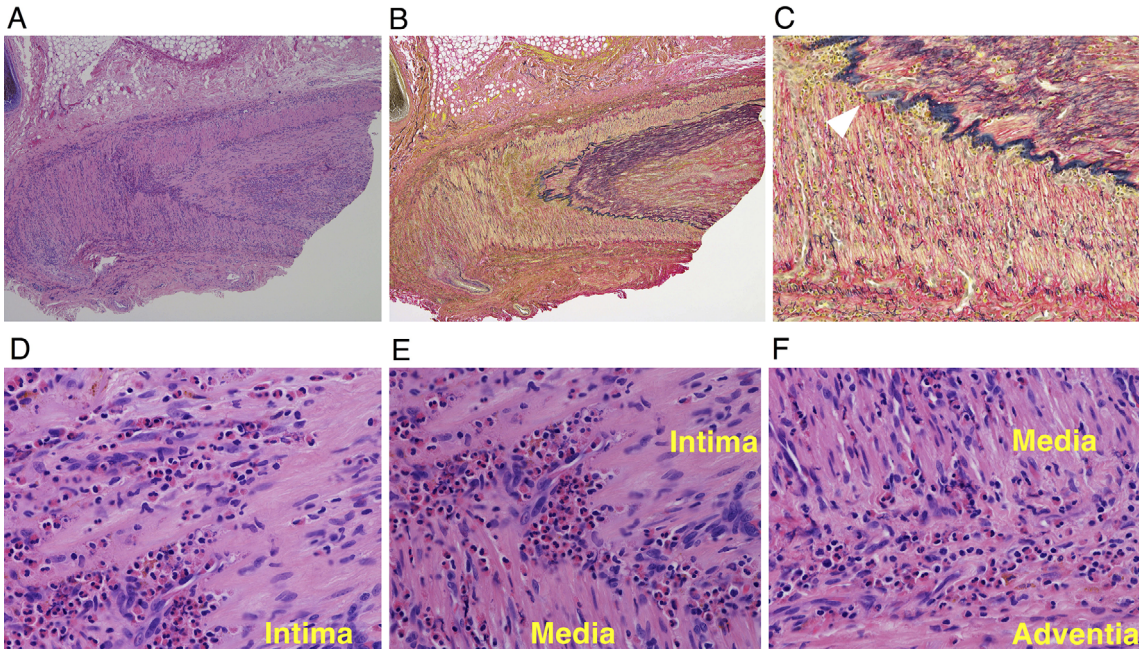

Figure 4. **A temporal artery biopsy specimen showing intense transmural infiltration of inflammatory cells, mainly composed of eosinophils in the absence of giant cell formation, and accompanied by disruption of the internal elastic lamina. A, and D-F: Hematoxylin and Eosin staining. B and C: Elastica van Gieson (EVG) staining. The original magnification is indicated in parentheses. A and B: A diagonal cross-section of the temporal artery at a lower magnification power (×40). C: Disruption of the internal elastic lamina is indicated by a white arrow (×200). Eosinophilic infiltration in the intima (D), intima-media junction (E), and media-adventia junction (F) is shown (×400), indicating transluminal distribution.**

TAO affecting the temporal arteries, and HES-associated vasculitis.

On her first visit to our hospital, the enlargement of the temporal arteries suggested the possibility of GCA, which is reported to involve upper extremities in approximately 10-30% of patients (34-40). However, the upper-limb involvement had resulted in digital ischemia in this case, which is uncommon in patients with GCA (41-44). Furthermore, the temporal artery biopsy specimen did not show granulomatous giant cell formation but demonstrated intense transmural inflammation mainly composed of eosinophils. Since the histological pattern of temporal artery biopsy specimens is discontinuous in 8-28% of cases of biopsy-proven GCA, known as skip lesions (45), the absence of giant cell formation does not preclude the possibility of GCA (46). However, dense eosinophil infiltration is an atypical finding in the diagnosis of GCA (47). Another differential diagnosis was JTA, which was first described by Lie et al. (48), as a rare inflammatory disease of the temporal arteries that affects young adults in the absence of systemic vasculitis. Journeau et al. recently summarized a total of 44 patients with JTA, including 12 patients who were newly identified by a multicentric survey in France (14). It is worth noting that their study showed the presence of eosinophilia in 15 of 44 patients with JTA (14). In this regard, Fujimoto et al. had already suggested that this subtype of JTA could be a distinct disease entity, which they named JTAE (49). However, given that our patient was 60 years of age, the presentation did not fit the criteria of JTAE. Furthermore, this case was complicated by upper-limb artery occlusion. Basically, JTAE is a localized vasculitis syndrome with eosinophilic infiltration confined to the temporal arteries. Therefore, the classification of JTAE would be inappropriate in this case.

Eosinophils are considered to play a key role in the pathogenesis of EGPA, which belongs to antineutrophil cytoplasmic antibody-associated small vessel vasculitis and which is characterized by asthma and other allergic symptoms, tissue and blood eosinophilia, and necrotizing granulomatous vasculitis. The involvement of temporal arteries by eosinophilic vasculitis with (50) or without giant cell formation (51-55) has been reported in patients with EGPA. MacDiarmid et al. reported a case of EGPA complicated by radial artery occlusion at the wrist-level (55). However, the patient in the current case had no history of asthma or other allergic diseases, lung involvement, or granulomatous giant cell formation on temporal artery biopsy, and was negative for Myeloperoxidase anti-neutrophil cytoplasmic antibody, which indicated that EGPA was an unlikely diagnosis. It should be noted here that she was a smoker and that US showed corkscrew-like deformity of the digital arteries. A corkscrew collateral appearance is a characteristic angiographic finding in patients with TAO (56). Furthermore, Lie et al. described 3 cases of TAO with eosinophilia involving the temporal arteries (51). Corticosteroid-responsiveness

does not preclude the possibility of TAO, since Naito et al. reported the case of a patient with TAO and blood hypereosinophilia who responded to corticosteroid treatment (19). However, in this case, angiography failed to show the typical angiographic findings of TAO, such as segmental occlusive lesions of the small and medium-sized vessels with the formation of distinctive small collateral vessels around the areas of occlusion (57). It should also be noted that several case reports have demonstrated that IgG4-related diseases (IgG4-RDs) can affect the temporal arteries with the marked infiltration of the adventitia by eosinophils and plasma cells (58, 59). However, in the present case, few plasma cells were observed in the temporal artery biopsy specimen, which excluded IgG4-RDs as a strong differential diagnosis.

The remaining possibility is eosinophilic vasculitis secondary to HES. Although according to the classical definition by Chusid et al., HES is indicated by AEC >1,500/mm^3 for >6 months (60), this requirement is infrequently applied today because of the need to start effective treatment early on in some patients to avoid irreversible organ damage (61). Thus, we consider that the present case-in which the AEC was >1,500/mm^3 without apparent causes-can be regarded as HES. Previous studies have demonstrated that HES can cause digital ischemia either by microthrombi (62, 63), or by the occlusion of intermediate to large-sized arteries (3, 4, 6, 11). Notably, a 28-year-old Japanese man with HES presenting with eosinophilic cellulitis, Raynaud's phenomenon, digital gangrene, and JTA was described by Ito et al., who proposed that JTAE may be one of the features of HES (10). Motegi et al. also suggested the concept of eosinophilic peripheral arteritis, defined as eosinophilia with peripheral arterial occlusion by eosinophilic arteritis in the extremities and temporal arteries as a part of the clinical manifestations of HES (29). Additionally, Alzayer et al. recently described the case of a 39-year-old patient who developed left hand digital ischemia, preceded by Raynaud's phenomenon, and vasculitic rash. They showed eosinophilic vasculitis without respiratory or renal involvement with the distal left ulnar artery and the radial artery branches in the left hand showing a beaded appearance on magnetic resonance angiography (2). Moreover, they proposed that hypereosinophilic vasculitis without respiratory or renal involvement is a distinct disease entity of the primary vasculitis that has not been properly recognized in the Revised International Chapel Hill Consensus Conference 2012 (CHCC 2012) classification system (12). The further accumulation of similar cases is warranted to characterize this rare type of eosinophilic vasculitis that affects the peripheral arteries.

The present study was associated with some limitations. We did not perform a histological examination of the proper palmar digital arteries in which cork-screw deformities were observed on Doppler US. Thus, we cannot clarify which of the two mechanisms, thrombosis formed by eosinophils or vascular damage due to eosinophilic vasculitis, contributed to the digital ischemia in this case. However, artery biopsy is not routinely performed in the clinical setting in cases similar to ours as it is considered to lead to the exacerbation of digital ischemia. The negative results of the coagulation-fibrinolysis system (D-dimers and FDP) suggest that eosinophilic thrombosis was less likely to have been involved in the development of peripheral ischemia, although the possibility cannot be completely excluded. It should also be noted that eosinophilia can be secondary to a large spectrum of underlying causes, including infections such as parasitic infections, allergies, autoimmune and inflammatory diseases, pulmonary eosinophilic disorders, adrenal insufficiency, and hematologic and solid neoplasms (64). Another limitation of the present case was that we did not perform molecular studies including screening for FIP1 L1-PDGFRA gene rearrangement, which should have been performed prior to corticosteroid treatment to exclude clonal eosinophil expansion. However, we consider the possibility for clonal eosinophil expansion to be low, since hypereosinophilia and the relevant clinical manifestations were well-controlled by low-dose corticosteroid treatment.

In conclusion, this report describes the case of an elderly woman with peripheral blood eosinophilia who developed non-granulomatous eosinophilic vasculitis of the temporal arteries complicated by digital ischemia, which was ameliorated by corticosteroid treatment. We consider that it is appropriate to classify the present case as eosinophilic vasculitis secondary to HES. However, eosinophilic vasculitis other than EGPA is not adequately incorporated in the CHCC2012 classification system. Thus, the classification system does not include the following eosinophilic scenarios in appropriate categories: recurrent cutaneous necrotizing eosinophilic vasculitis (65-68), cutaneous eosinophilic necrotizing vasculitis secondary to connective tissue disease (69, 70), JTAE, and eosinophilic vasculitis affecting the relatively medium-sized vessels of the limbs that is distinctive from cutaneous eosinophilic vasculitis (2-11). Further studies to characterize the clinical manifestations of these eosinophilic vasculitis syndromes are necessary to establish more comprehensive nomenclature and classification of vasculitis.

The authors state that they have no Conflict of Interest (COI).

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