Case Report

Caught between Two Storms: A Rare Vasculitis with Complicated Infections

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Abstract

Eosinophilic granulomatosis with polyangiitis (EGPA) is a rare mini- to average-sized vessel necrotizing vasculitis marked by asthma, eosinophilia, and systemic involvement. Diagnostic challenges arise due to its varied presentations and low rate of antineutrophil cytoplasmic antibody (ANCA) positivity. Here, we present the case of a 69-year-old male with chronic bronchitis and recent nasal surgery for rhinosinusitis. Postoperatively, the patient had an ischemic stroke, where embolic sources were excluded, and treated conservatively, and is now presented to us with progressive distal limb weakness—mononeuritis multiplex. Furthermore, evaluation, laboratory tests showed significant eosinophilia (65%) with positive p-ANCA and antinuclear antibodies, started on steroid therapy. Meanwhile, the patient deteriorated with severe oronasal bleeding with hemodynamic compromise, shifted to the intensive care unit, and was initiated on invasive mechanical ventilatory and hemodynamic support. Bronchoscopy and endoscopic cauterization of bleeding vessels were done, but we could not initiate the patient on immunosuppressant therapy due to concurrent tuberculosis confirmed via bronchoalveolar lavage with cultures positive for methicillin-resistant *Staphylococcus aureus* infection. Plasma exchange was initiated due to suspected active vasculitis, and the patient is slowly weaned off all support. EGPA with active vasculitis presenting as a bleeding emergency with complicated coinfections is the first case reported to date.

Keywords: *Antineutrophil cytoplasmic antibody, antinuclear antibodies, Echurg-Strauss syndrome, eosinophilia, eosinophilic granulomatosis with polyangiitis, methicillin-resistant Staphylococcus aureus, plasma exchange, tuberculosis*

Introduction

Eosinophilic granulomatosis with polyangiitis (EGPA), previously termed Churg–Strauss syndrome, is an uncommon systemic necrotizing vasculitis. It is clinically presented with adult-onset asthma, peripheral blood eosinophilia, and chronic rhinosinusitis.[1] EGPA is categorized under antineutrophil cytoplasmic antibody (ANCA) associated vasculitis; however, ANCA positivity is detected in only 30%–40% of cases,[2] and it involves various organ systems, making it a multisystem disease,[3] with varied presentations, complicating the diagnostic process. Each year, approximately 0.5–4.2 cases per million are reported, although this figure may be lower than the actual incidence due to underdiagnosis of EGPA.[1]

Case Presentation

A 69-year-old male with chronic bronchitis and a recent history of nasal polypectomy for rhinosinusitis experienced a postoperative ischemic stroke without lasting deficits and later presented with left upper limb sensory disturbances lasting 10 days and decreased hand grip strength, loss of sensation over the dorsum of the hand, difficulty walking, and foot drop. Magnetic resonance imaging of the brain demonstrated a new acute lacunar infarct in the right peritrigonal white matter. Computed tomography (CT) neck angiography and transesophageal echocardiography were unremarkable. Bubble contrast echocardiography and 24-h Holter monitoring showed no evidence of an embolic source. Laboratory evaluation revealed marked eosinophilia (65%). In view of mononeuritis multiplex and esinophillic leukocytosis, further evaluation was done with antinuclear antibodies, which turned out to be positive with a nucleus speckled pattern as shown in Figure 1. P-ANCA was positive. Planned for sural nerve biopsy and pulse steroid therapy. Meanwhile, the patient

How to cite this article: Shilpa G, Sheshala K, Bhowmick N, Surisetti BK, Ugandhar Bhattu C, Sudhakar G. Caught between two storms: A rare vasculitis with complicated infections. J Med Res 2026;12:84-7.

Received: 22-Jan-2026 **Revised:** 23-Apr-2026 **Accepted:** 07-May-2026 **Web Publication:** 05-Jun-2026

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Access this article online

Website: https://journals.lww.com/jmer

DOI: 10.4103/JMR.JMR_7_26

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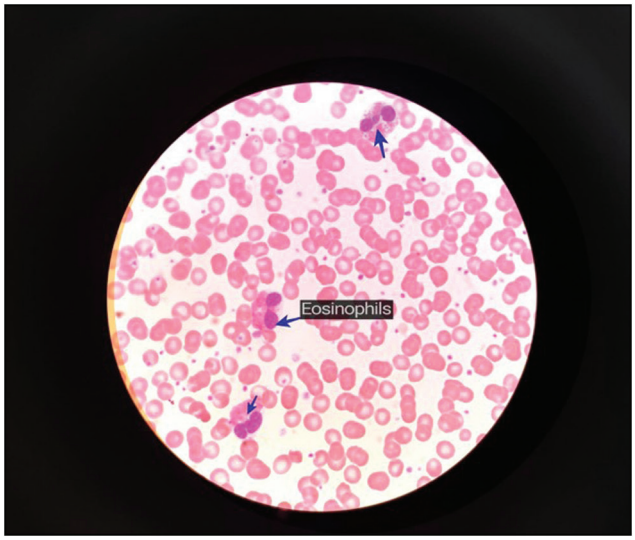

Figure 1: Antinuclear antibodies that turned out to be positive with a nucleus speckled pattern

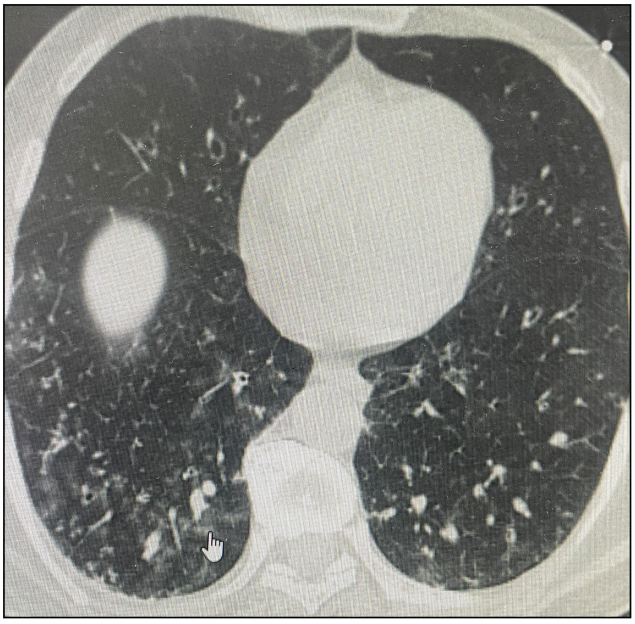

Figure 2: Computed tomography of the chest suggestive of bilateral ground-glass opacities

had a severe oronasal bleed, hence the patient was shifted to the intensive care unit (ICU). Initial resuscitation was done with multiple blood products, and electively intubated for airway protection. In view of worsening hemodynamics, vasopressor support was started. CT of the chest suggestive of bilateral ground-glass opacities, as shown in Figure 2. Pulse dose steroid (injection methylprednisolone 1 g for 3 days) was given, Bronchoscopy suggestive of hyperemic mucosa with presence of hemosiderin-laden macrophages (HLMs) and broncho alveolar XPERT mycobacterium tuberculosis MTB, cartridge based nucleic acid amplification test detected tuberculosis (TB), broncho alveolar (BAL) cultures grew methicillin-resistant *Staphylococcus aureus* (MRSA).

Patient continued to have a nasal bleed, and resuscitation was continued. In view of hemodynamic instability, the patient was directly taken to occupational therapy for endoscopic exploration, and an active spurt from ethmoidal and bilateral sphenopalatine arteries was noted. Cauterization of the bilateral sphenopalatine artery was continued on mechanical ventilatory support. In view of active vasculitis (arteritis) patient need of aggressive immunosupressants was discussed as steroid therapy alone would not suffice; meanwhile, BAL analysis reported TB positive status with MRSA infection, active immunosupressants were deffered and went ahead with plasma exchange, total four sessions were done, started on anti tubercular therapy and targeted antibiotics, infection and active vasculitis was controlled, patient weaned from all supports and discharged home with improvement in neurologic symptoms.

Discussion

Most patients exhibit a typical three-phase clinical pattern, as illustrated in Figure 3; however, EGPA does not always follow a strict or sequential progression. The phases may overlap or be incomplete, with some individuals showing only selected features. The transition from the initial allergic phase to the vasculitic stage usually occurs gradually with an average duration ranging from 3 to 9 years.[4]

ANCA-associated vasculitides present challenges due to their diverse clinical features. Several classification and diagnostic criteria have been developed to improve recognition, including the 1984 Lanham criteria, the American College of Rheumatology guidelines, the 2012 revised Chapel Hill Consensus definitions, and the EGPA Consensus Task Force recommendations, supporting identification and clinical trials.[5]

The 2022 American College of Rheumatology European Alliance of Associations classification criteria represent a validated instrument to aid in diagnosing EGPA. It is intended for patients with an established small- or average-sized vessel vasculitis diagnosis. The system assigns weighted scores to individual criteria, and a total score of 6 or higher supports classification of EGPA [Table 1].

Due to the multisystem involvement of EGPA, the patient's presenting complaints guided the diagnostic approach undertaken. In this case report, foot drop accompanied by paresthesias and multiple neurological findings on examination initially suggested a primary neurological cause. Neurological manifestations in EGPA are closely associated with eosinophilia, which contributes to axonal neuropathy.[2] This process is believed to result from neurotoxic substances released by infiltrating eosinophils, leading to damage of adjacent nerve fibers. Furthermore, neurological involvement is frequently linked with ANCA positivity,[4] which was also observed in this patient. Such overlapping features often complicate diagnosis and require careful clinical and laboratory correlation evaluation.

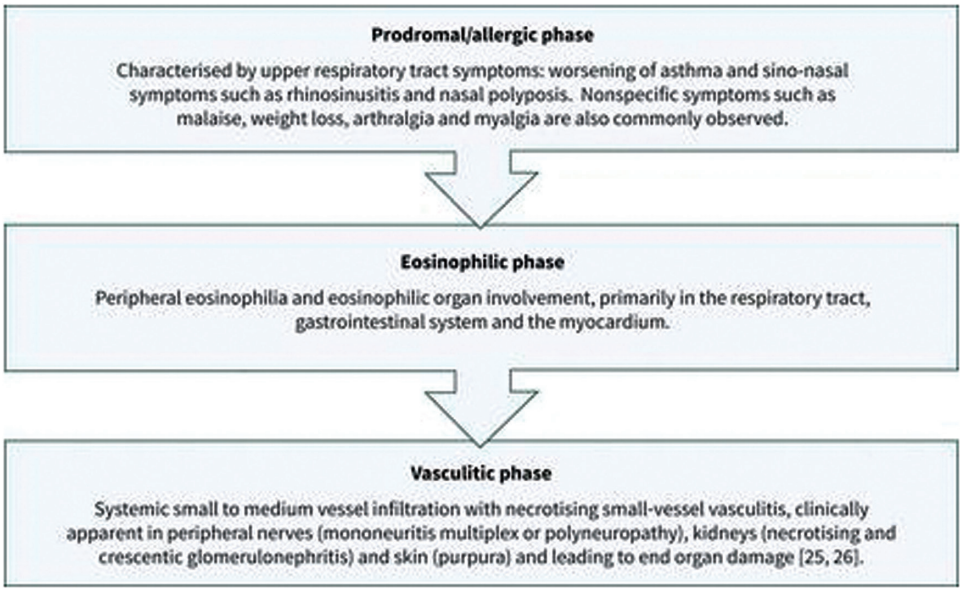

Figure 3: Three clinical phases of eosinophilic granulomatosis with polyangiitis

Table 1: The 2022 American College of Rheumatology/ European Alliance of Associations eosinophilic granulomatosis with polyangiitis classification criteria

Criteria Type	Condition	Intensity
Clinical criteria	Nasal polyps	+3
	Obstructive airway disease	+3
	Mononeuritis multiplex	+1
Laboratory criteria	Blood eosinophil count ≥1 × 109 cells/L	+5
	Biopsy evidence of extravascular eosinophilic inflammation	+2
	Positive test for cytoplasmic antineutrophilic cytoplasmic antibodies or anti-proteinase 3 antibodies	−3
	Hematuria	−1

However, the severe active vasculitis with unexpected bleeding emergency was atypical in our case, which even required endoscopic cauterization.

BAL fluid showed the presence of HLMs with XPERT MTB, and CBNAAT detected TB, with cultures growing MRSA, making management of this patient more complicated, as immunosuppressants for active vasculitis may worsen the underlying infection. The patient underwent plasma exchange, and eventually improved after four sessions of plasma exchange.

Conclusion

This case report details a rare, complex, and high-risk presentation of p-ANCA-positive EGPA. The patient's initial manifestation with mononeuritis multiplex, followed by a catastrophic hemorrhagic emergency, the clinical course was further complicated by a concurrent diagnosis of active TB with MRSA coinfection, which precluded the use of standard immunosuppressive therapies and necessitated an innovative management approach.

This case report highlights key clinical insights, emphasizing that EGPA, although uncommon, must be included in the differential diagnosis when patients present with features suggestive of mononeuritis multiplex, in the context of adult-onset asthma, rhinosinusitis, and peripheral eosinophilia, particularly when common cardioembolic sources have been excluded. This case report provides a successful management paradigm for a patient with potentially fatal ANCA-associated vasculitis and a contraindication to conventional immunosuppression, demonstrating that plasma exchange can serve as a highly effective and life-saving bridging therapy. The reporting of such complex and atypical cases is essential for expanding the collective clinical understanding of the full spectrum of EGPA and for refining our strategies to manage its most challenging presentations.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Author Contributions

GPS, Dr. KS, NB, BKS, CUB, GS conceptualized, designed and supervised the study. GPS and KS did the literature search and data acquisition. All the contributors were involved in conducting the clinical and experimental studies. Manuscript preparation was done by GPS and KS. Manuscript editing was done by all authors. All authors

reviewed and approved the final version of the manuscript. GPS acts as a Guarantor.

Data Availability Statement

The data supporting the findings of this study can be obtained from the corresponding author upon request.

Use of AI in drafting the manuscript

The authors declare that they have not used any generative AI/AI-assisted technologies in the writing of this manuscript.

Financial support and Sponsorship

Nil.

Conflicts of Interest statement

There are no conflicts of interest.

Acknowledgement

We would like to thank Management Yashoda Hospitals and Dr. Amidayala Lingaiah (Director -Medical Services) for the continuous support.

Ethical Aspects

We state that the manuscript has been read and approved by all the authors, the requirements for authorship as stated earlier in this document have been met, and each author believes that the manuscript represents honest work.

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