

Rapid Clinicoradiologic Remission of Relapsing Idiopathic Chronic Eosinophilic Pneumonia with Benralizumab: A Case Report

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ABSTRACT

Idiopathic chronic eosinophilic pneumonia (ICEP) is an eosinophil-driven interstitial lung disease with brisk corticosteroid responsiveness but frequent relapses that increase cumulative steroid exposure.

A 36-year-old woman with a 13-year history of relapsing ICEP (≥ 4 steroid-responsive flares) presented with cough and exertional dyspnea. Baseline eosinophils were $1.74 \times 10^3/\mu\text{L}$ (17%), CRP 52.2 mg/L, and ESR 79 mm/h; chest radiograph and HRCT showed bilateral, predominantly peripheral consolidations. Off-label benralizumab 30 mg s.c. was administered without systemic steroids. By day 15, the chest radiograph showed near-complete clearing; eosinophils fell to 0%, CRP to 9.47 mg/L, and ESR to 31 mm/h.

Benralizumab was associated with a rapid early hematologic and radiologic response and allowed avoidance of systemic corticosteroid use during the index relapse in relapsing ICEP.

Keywords: Carrington syndrome, benralizumab, interleukin-5 receptor alpha subunit, pulmonary eosinophilia, eosinophils

INTRODUCTION

Idiopathic chronic eosinophilic pneumonia (ICEP), also known as Carrington syndrome, presents subacutely with cough/dyspnea, blood or BAL eosinophilia, and peripheral, non-segmental consolidations—classically described as the “photographic negative of pulmonary edema” (1, 2). Although corticosteroids usually induce rapid clinical and radiological improvement, relapses are common during dose tapering or after treatment withdrawal, leading to cumulative corticosteroid exposure and toxicity (2). Differentiation from acute eosinophilic pneumonia (AEP) is essential because AEP typically presents as an acute hypoxemic syndrome with different clinical and radiological features (3). Given the central role of eosinophils in disease pathobiology, biologics targeting interleukin-5 (IL-5) or its receptor alpha subunit (IL-5R α), particularly benralizumab, are increasingly being reported as steroid-sparing options in relapsing ICEP (4-7).

CASE REPORT

A 36-year-old never-smoking woman had been followed for ICEP since 2012 with ≥ 4 documented relapses. Each relapse responded promptly to prednisone 16 mg twice daily for 3 months, but symptoms recurred after discontinuation. She presented with cough and exertional dyspnea; she was afebrile with room-air SpO₂ 92-93% and bilateral fine crackles.

Baseline laboratory tests: leukocytosis with absolute eosinophils $1.74 \times 10^3/\mu\text{L}$ (17%), CRP 52.2 mg/L, ESR 79 mm/h.

Imaging: Pre-treatment postero-anterior chest radiograph demonstrated bilateral, predominantly peripheral consolidations (Figure 1). HRCT showed patchy peripheral and upper-lobe-predominant consolidations and ground-glass opacities, a pattern compatible with ICEP and distinct from AEP (Figure 3A).

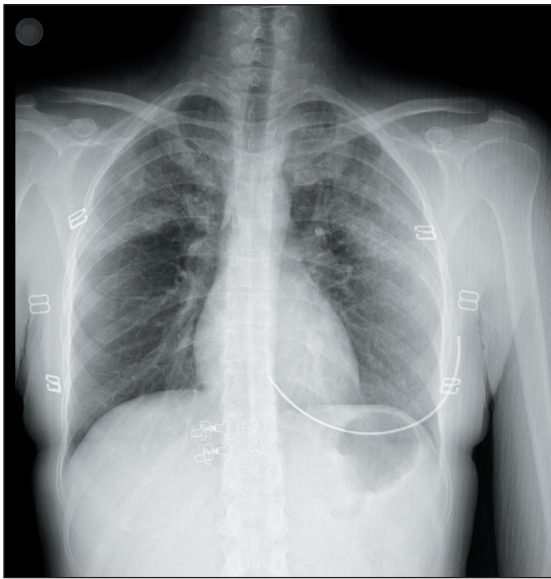


Figure 1: Pre-treatment postero-anterior chest radiograph showing bilateral peripheral consolidations consistent with active ICEP.

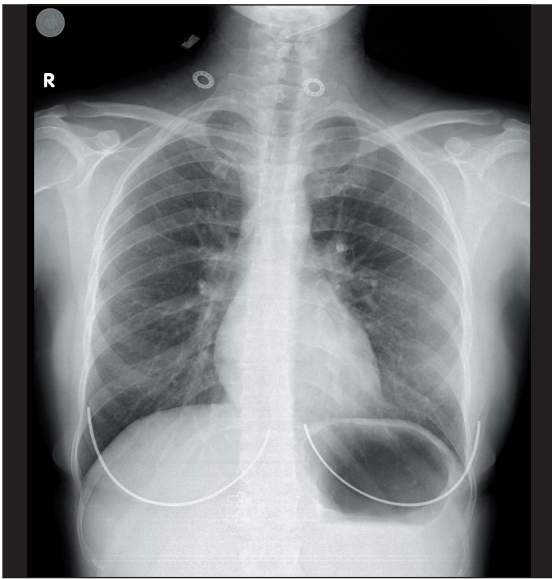


Figure 2: Follow-up postero-anterior chest radiograph obtained on day 15 after benralizumab, demonstrating near-complete clearing of the previous peripheral opacities.

Table I: Key laboratory and follow-up findings

Parameter	Baseline	Day 15 / Follow up
Eosinophils, %	17	0.0
Eosinophils, $\times 10^3/\mu\text{L}$	1.74	0.00
CRP, mg/L	52.2	9.47
ESR, mm/h	79	31
SpO ₂ , %	92-93	97
Bronchoalveolar lavage	Not performed during the index relapse	-
ANCA	Not available during the index relapse	-
Follow-up HRCT timing	-	1 month

Treatment and monitoring: Given the relapsing, steroid-dependent course and the aim to limit further systemic steroid exposure, off-label benralizumab 30 mg subcutaneously was initiated (severe eosinophilic asthma schedule planned: weeks 0, 4, 8, then every 8 weeks). Inhaled controller therapy was continued; no systemic steroids were started at this presentation. Written informed consent for off-label therapy and publication was obtained.

Outcomes: By day 15, cough and breathlessness markedly improved; room-air SpO₂ increased to 97%. A follow-up postero-anterior chest radiograph demonstrated near-

complete clearing of the previous peripheral opacities (Figure 2). The eosinophil count fell to 0% ($0.00 \times 10^3/\mu\text{L}$); CRP decreased to 9.47 mg/L and ESR to 31 mm/h (Table I). A follow-up HRCT obtained at 1 month showed substantial regression of the previously observed peripheral consolidations and ground-glass opacities, with minimal residual subpleural changes (Figure 3B).

DISCUSSION

This case displays the classic ICEP clinico-radiologic pattern—subacute course, eosinophilia, and peripheral/upper-lobe-predominant consolidations—contrasting with AEP’s acute hypoxemic presentation (1-3). Benralizumab binds the interleukin-5 receptor alpha subunit on eosinophils and induces near-complete eosinophil depletion through antibody-dependent cellular cytotoxicity, providing a strong biological rationale for its use in eosinophilic lung diseases (4). Published case reports and recent reviews suggest that anti-IL-5 or anti-IL-5 receptor therapies may induce rapid symptom control, radiological improvement, and corticosteroid reduction in relapsing ICEP (5-7). In several reported patients, improvement has been observed within days to weeks after treatment initiation, and maintenance schedules have generally followed severe eosinophilic asthma regimens, although the optimal duration of therapy remains uncertain (4, 7). In our patient, benralizumab was associated with marked im-

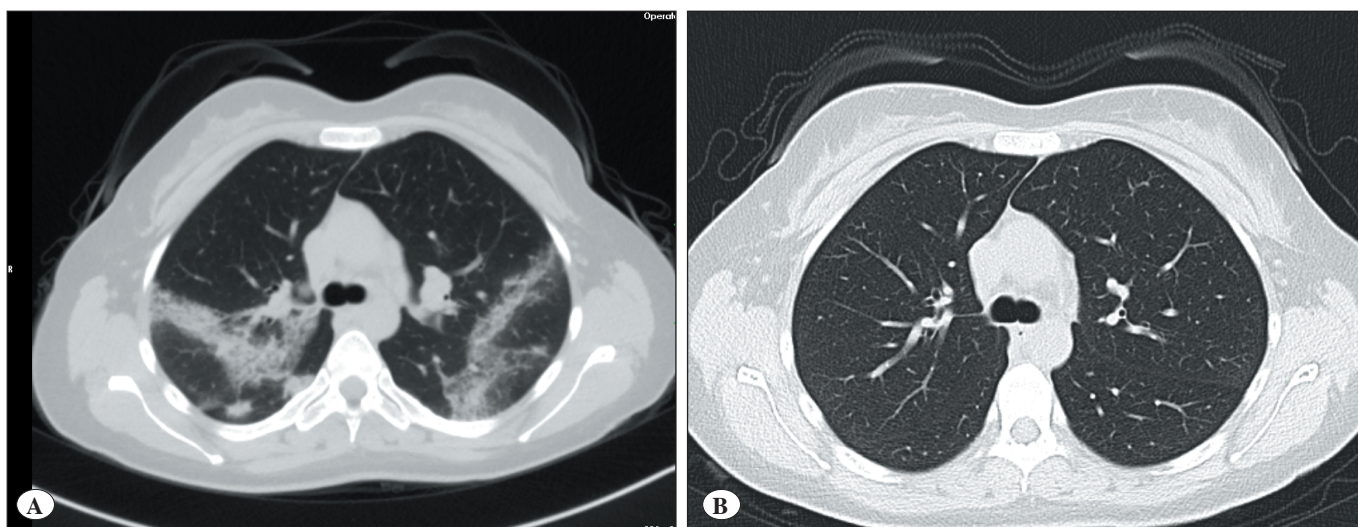


Figure 3: **A)** Pre-treatment high-resolution computed tomography (HRCT) image showing peripheral and upper-lobe-predominant consolidations with ground-glass opacities. **B)** Follow-up high-resolution computed tomography (HRCT) image obtained at 1 month after benralizumab, demonstrating marked regression of the previously observed consolidations and ground-glass opacities, with minimal residual subpleural changes.

provement by day 15 in symptoms, blood eosinophils, inflammatory markers, and chest radiography, while follow-up HRCT at 1 month confirmed substantial radiological regression.

Limitations

This report has several limitations. First, it describes a single case, which limits generalizability. Second, follow-up is currently limited and therefore does not allow conclusions regarding long-term disease control. Third, bronchoalveolar lavage was not performed and ANCA testing was not available during the index relapse. Therefore, although the diagnosis was strongly supported by the patient's previously established relapsing idiopathic chronic eosinophilic pneumonia, characteristic radiological pattern, and marked peripheral eosinophilia, the present report should be interpreted as documenting a rapid early clinoradiologic response rather than a fully comprehensive diagnostic re-evaluation of the flare. Finally, the steroid-sparing effect was not assessed using a predefined cumulative corticosteroid reduction model; accordingly, this case is best interpreted as demonstrating avoidance of systemic corticosteroid use during the index relapse.

CONCLUSION

In this patient with relapsing idiopathic chronic eosinophilic pneumonia, benralizumab was associated with a rapid early clinoradiologic response and avoidance of systemic corticosteroid use during the index relapse. Further studies are needed to clarify the optimal duration and long-term role of anti-IL-5/IL-5 receptor therapy in ICEP.

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Ethical Approval and Patient Consent

Institutional review board approval was not required for a single anonymized case report according to local policy. Written informed consent was obtained from the patient for publication of anonymized clinical data and images.

Conflict of Interest

The authors declare no competing interests.

Author Contributions

Concept: MHB, Design: MHB, Data collection or processing: MHB, AA, Analysis or Interpretation: MHB, AA, Literature search: MHB, AA, Writing: MHB, Approval: MHB, AA.

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