

Mepolizumab for the treatment of eosinophilic cystitis: reply

Trefond L^{1,2} Kahn JE³

¹Service de Médecine Interne, CHU Gabriel Montpied, Clermont-Ferrand, France

²Université Clermont Auvergne, Inserm U1071, M2iSH, USC-INRA 1382, ClermontFerrand, France

³National Reference Center for Hypereosinophilic Syndromes, CEREO, France; Université Paris-Saclay, Assistance Publique - Hôpitaux de Paris, Department of Internal Medicine, Ambroise Paré Hospital, Boulogne-Billancourt, France

Corresponding author:

Ludovic Trefond

Université Clermont Auvergne, Inserm U1071, M2iSH, USC-INRA 1382

Service de Médecine Interne, CHU Gabriel Montpied Clermont Ferrand

63000 ClermontFerrand, France

E-mail: ltrefond@chu-clermontferrand.fr

This article has been accepted for publication and undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process, which may lead to differences between this version and the Version of Record. Please cite this article as doi: 10.18176/jiaci.0980

Key words: Hypereosinophilic syndrome. Eosinophilic cystitis. Mepolizumab.

Palabras clave: Síndrome de hipereosinofílico. Cistitis eosinofílica. Mepolizumab.

To the Editor,

We read with interest the publication by Wang and colleagues on the off-label use of Mepolizumab for the treatment of eosinophilic cystitis in a 77-year-old patient [1]. The authors presented another patient with idiopathic eosinophilic cystitis successfully treated with mepolizumab [1]. We appreciate the authors' contribution in reference to our publication on the first two cases of eosinophilic cystitis (EC) successfully treated with mepolizumab [2].

In Wang et al.'s reported case, there was notably a biological inflammatory syndrome initially, with a CRP of 40 mg/L and an ESR of 25 mm/h, while in the literature review of 135 patients with EC in 2000 [3], only 7% of patients had an elevated ESR. Our two patients [2] did not exhibit an inflammatory syndrome. We also lack details on the eosinophil infiltration rate in the biopsy in Wang et al.'s case. Our patients had counts of 200/HPF and 180/HPF, significantly higher than the cutoff of 15/HPF in eosinophilic esophagitis [4]. Another surprising aspect is the absence of imaging abnormalities in Wang et al.'s patient [1], whereas in our cases, imaging helped for diagnosis and provided clear evidence of improvement under treatment.

Despite these considerations, this case appears compelling and aligns with an organ-restricted presentation without HE, similar to our case 1 [2].

In Table 1, we have summarized the key elements from these three cases [1,2].

Despite the limited follow-up in Wang et al.'s case [1], there have been no reported relapse. This supports the notion, as we previously suggested, that mepolizumab may hold promise in the off-label treatment of idiopathic eosinophilic cystitis.

Funding

The authors declare that no funding was received for the present study.

Conflicts of Interest

J.-E. Kahn reports consulting fees for advisory boards from AstraZeneca and GSK, research funding from AstraZeneca and GSK, and participation in clinical trials sponsored by AstraZeneca. The remaining authors declare no conflict of interest.

References

1. Gang W, Ning Z, Zhichun L. Off-label Use of Mepolizumab: A Potential Therapeutic Option for Eosinophilic Cystitis. *J Investig Allergol Clin Immunol*. Epub ahead of print.
2. Trefond L, Guy L, Darcha C, Gallon A, Thomas-Monier R, Berdugo K, Smets P, Olagne L, Stievenart J, Fayard D, Cathebras P, Aumaitre O, Boyer L, Andre M, Kahn JE. Efficacy of mepolizumab for the treatment of eosinophilic cystitis: a report of two cases. *J Investig Allergol Clin Immunol*. 2023 Oct 17:0. doi: 10.18176/jiaci.0954. Epub ahead of print. PMID: 37850410.
3. Van den Ouden D. Diagnosis and management of eosinophilic cystitis: a pooled analysis of 135 cases. *Eur Urol*. 2000 Apr;37(4):386–94.
4. Attwood S, Epstein J. Eosinophilic oesophagitis: recent advances and practical management. *Frontline Gastroenterology*. 2021 Dec 1;12(7):644–9.

Table 1. Characteristics of three cases of eosinophilic cystitis successfully treated by off-label use of Mepolizumab.

	Case 1 *	Case 2 *	Case 3 **
Country	France	France	China
Age (years)	69	15	77
Symptoms	Hematuria, dysuria, urinary frequency	Abdominal pain, pollakiuria, dysuria	Difficulty in urination, frequent urination
Eosinophilia (x10 ⁹ /L)	660	3000	850
IgE	1500kU/L	1800 kU/L	175UI/mL
CRP (mg/L)	<5	<5	40
Eosinophiluria	90 eo/100 cells		
Biopsy	200/HPF	180/HPF	Eosinophilic infiltration
Treatment before Mepolizumab	Prednisone	Prednisone	Methylprednisolone and Tripterygium wilfordii
Other organ	-	Eosinophilic gastroenteritis	-
Type of HES ***	Organ-restricted without HE	Multi-organ involvement with severe HE	Organ-restricted without HE
Relapse	-	-	-
Follow up	1 year	15 years	6 months

CRP, C-reactiv protein, HE hypereosinophilia, HES hypereosinophilic syndrome

* Trefond L, Guy L, Darcha C, Gallon A, Thomas-Monier R, Berdugo K, Smets P, Olgne L, Stievenart J, Fayard D, Cathebras P, Aumaitre O, Boyer L, Andre M, Kahn JE. Efficacy of mepolizumab for the treatment of eosinophilic cystitis: a report of two cases. *J Investig Allergol Clin Immunol*. 2023 Oct 17:0. doi: 10.18176/jiaci.0954. Epub ahead of print. PMID: 37850410.

** Gang W, Ning Z, Zhichun L. Off-label Use of Mepolizumab: A Potential Therapeutic Option for Eosinophilic Cystitis. *J Investig Allergol Clin Immunol*. Epub ahead of print.

*** Valent P, Klion AD, Roufousse F, Simon D, Metzgeroth G, Leiferman KM, et al. Proposed refined diagnostic criteria and classification of eosinophil disorders and related syndromes. *Allergy*. 2023 Jan;78(1):47–59