Difficult to treat relapsing polychondritis responded to tocilizumab in a teaching hospital in Western Kenya: Case report

Aksam R[1], Omondi C[1,2]

[1]Department of Medicine, Jaramogi Oginga Odinga Teaching and Referral Hospital, Kisumu, Kenya

[2]Department of Medicine, Uzima University, Kisumu, Kenya

Corresponding author:
Dr. Charles Omondi, Department of Medicine, Jaramogi Oginga Odinga Teaching and Referral Hospital, Kisumu, Kenya. Email: gomondii@yahoo.com

Abstract

Relapsing polychondritis is a rare autoimmune condition affecting the cartilaginous areas. Some cases respond poorly to conventional Disease Modifying Antirheumatic Agents (DMARDs) and may need biological agents. Here we present such as a case from a teaching hospital in Kisumu, Kenya. A 35 year old lady presented with a one year history of recurrent ear and nasal inflammation, eye pain and reddening, joint pains and swelling, and skin rash affecting the limbs and the trunk. She had been on treatment over the last one year with steroids, leflunomide, methotrexate and cyclophosphamide without significant improvement. Clinical exam showed: episcleritis, erythema nodusum-like skin rash, inflammation of the pinnae, bilateral ankle arthritis and saddleback deformity of the nose. Her inflammatory markers were elevated. She was started on monthly intravenous tocilizumab at 6mg/kg and made remarkable improvement. Tocilizumab is an effective therapy for difficult to treat relapsing polychondritis.

Key words: Relapsing polychondritis, Difficult, Treat, Tocilizumab, Responded

Introduction

Relapsing polychondritis is a rare autoimmune condition affecting the cartilaginous areas including the eyes, ears, nose, trachea, skin, joints among other areas. Majority of the cases respond to steroids and conventional DMARDs (cDMARDs). However, some cases are difficult to treat. Here we present such a case from Jaramogi Oginga Odinga Teaching Hospital (JOOTRH), Kisumu, Kenya who had been managed with steroids and conventional DMARDs for a year without improvement, but showed significant improvement on interleukin-6 (IL-6) blocker, tocilizumab.

Case report

A 35 year old African lady, married with three children presented to the clinic with a one year history of recurrent ankle joint pain and swelling, nodular skin lesion affecting the limbs and trunk, pain and reddening of the eyes, and nasal itchiness and deformity. She had no dyspnea or recurrent wheeze. There were no urinary or gastrointestinal symptoms. She does not smoke cigarettes. She had not experienced such symptoms before and neither did she have first-degree family history of similar symptoms. She had been on treatment for the past one year with multiple drugs including: methotrexate, leflunomide, cyclophosphamide, oral methylprednisolone without much improvement. Clinical exam showed: episcleritis of both eyes, bilateral ankle arthritis, erythema nodosum-like rash on the trunk and limbs, pinnae inflammation (Figure 1), sterile pustulosis of the hands, saddleback deformity of the nose (Figure 2). Cardiovascular, central nervous and gastrointestinal exam were found normal.

Her laboratory markers were as follows: ESR 30mm/hr, Hepatitis B surface antigen negative, Hepatitis C antibody negative, HIV antibody negative, urinalysis normal findings. Her creatinine at admission was 76umol/L, AST 26 IU/L, ALT 36 IU/L, serum albumin 46g/L, creatinine phosphokinase at 46IU/L. Her P-ANCA and C-ANCA were both negative. Her electrocardiogram and chest radiographs were normal. Her CT scan chest was normal (Figures 3, 4). She was started on monthly intravenous tocilizumab at 6mg/kg (total dose 400mg for her 66kg weight) for a total of six cycles and maintained on once daily prednisolone at 10mg while on tocilizumab (Figure 5). She made remarkable recovery with correction of saddleback nasal deformity (Figure 6). She was maintained on prednisolone 10mg once daily after tocilizumab.

Figure 1: Inflammation of the ear with pustular lesion

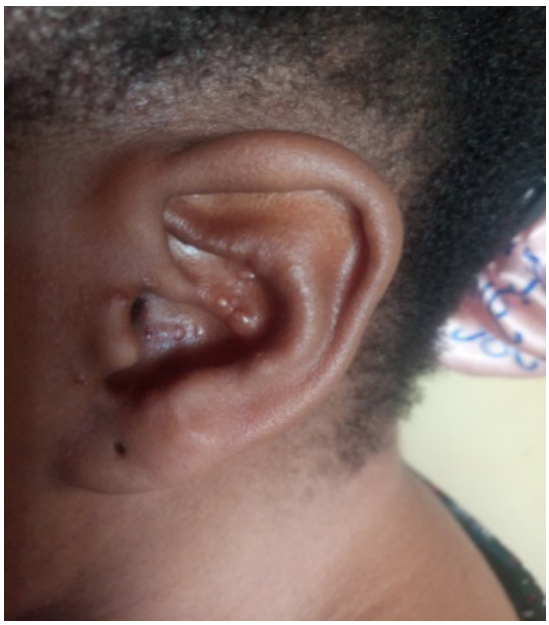


Figure 2: Saddleback nasal deformity at first presentation

Figure 3: Coronal view, normal CT scan chest

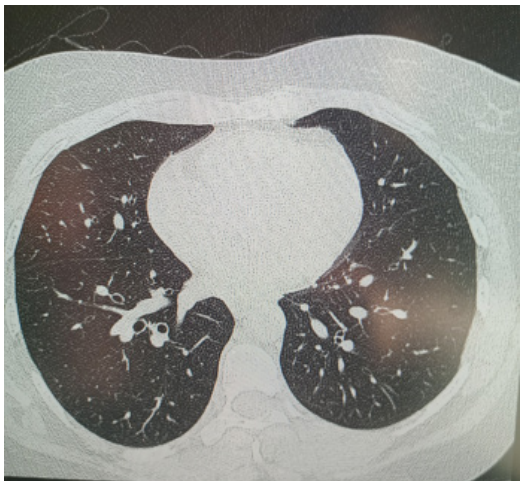


Figure 4: Sagittal view, normal CT scan

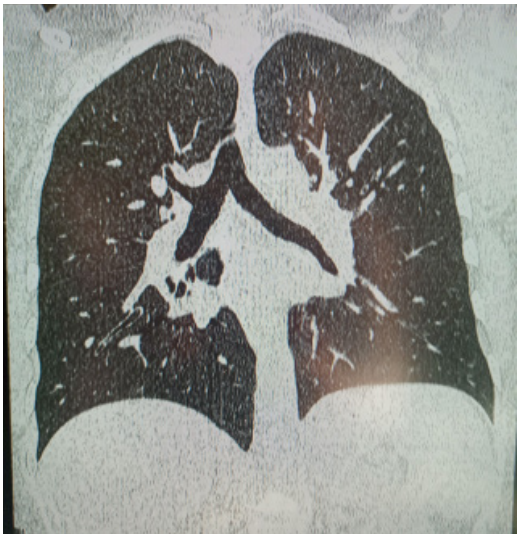


Figure 5: Improvement of ear inflammation with tocilizumab treatment

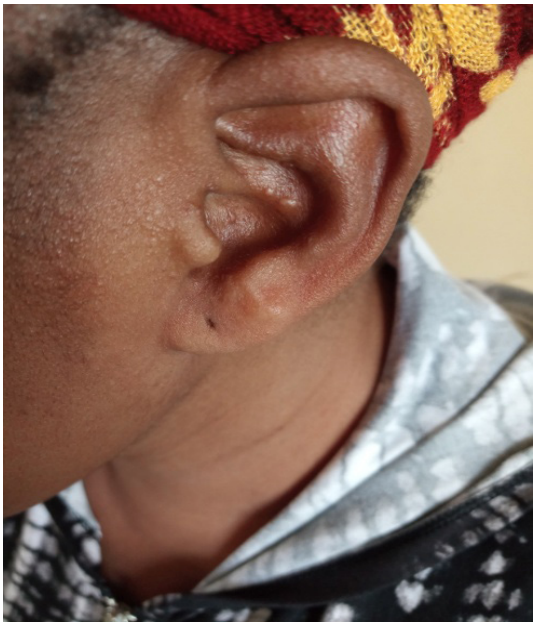
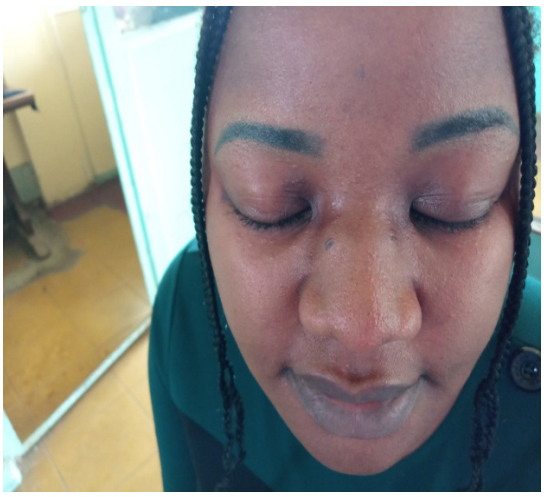


Figure 6: improvement of the saddleback nasal deformity with tocilizumab treatment



Discussion

This is the first recorded case of relapsing polychondritis managed by tocilizumab at JOOTRH. This case illustrates the efficacy of tocilizumab in the management of recalcitrant relapsing polychondritis after failure of both methotrexate and cyclophosphamide treatment. This compares well with other rheumatologists elsewhere in Europe, America and Asia who have successfully managed recalcitrant relapsing polychondritis with tocilizumab[1-3].

Despite spirited efforts to check the major scientific publication sources including Scorpio, Pubmed, Google scholar etc there were few reported case reports or case series from Africa where relapsing polychondritis had been managed by tocilizumab. Some publications reported cases of malignancy-associated relapsing polychondritis[4,5].

In conclusion**,** tocilizumab is an effective therapy for recalcitrant relapsing polychondritis.

Declaration

Conflict of interest: The authors declare no conflict of interest.

Drug donation: Novartis Kenya Ltd, drug initially donated for management of Covid-19 but remainder used in the management of rheumatic diseases.

Patient consent: Patient consent was sort and permission to publish granted.

Patient care: Both Dr. Rukia Aksam and Dr. Charles Omondi took care of the patient in the ward.

Radiological investigation: Provided by Dr. Zebedee Akanga, radiologist and Chair, Department of Radiology, JOOTRH, Kisumu, Kenya

References

1. Liao H. Efficacy of tocilizumab for refractory relapsing polychondritis with tracheal, respiratory distress. *Rheumatol.* 2021; **61:**(3): 1293-94.
2. Laimer T, Treiber M, von Werder A, *et al.* Relapsing polychondritis: an autoimmune disease with many faces. *Autoimmune Rev.* 2010; **9:**540-546.
3. Doctor M, Murthy SI, Rajusekhar L. Tocilizumab in recalcitrant bilateral scleritis in a case of relapsing polychondritis: A 17-year follow up. *Oral Immunol Inflamm.* 2023; **31**(4): 870-873.doi: 10.1080/09273948.2022.2058555.Epub 2022 June 13.PMID:35695904
4. Marshall J, Leroux DG. Chronic atrophic polychondritis: a South African case. *S Afr Med J.* 1964; **38**:527-529
5. Gning SB, Perret JL, Cissokho B, *et al.* Polychondrite atrophiante associee a` un adenocarcinome rectal et une fibromatose musculoaponevrotique chez une Africaine[Atrophic polychondritis associated with rectal adenocarcinoma and muscular aponeurotic fibromatosis in African woman]. *Rev Med Intern.* 2001; **22**(9): 891-893. French.doi:10.1016/s0248-8663(01)00444-1.PMID:11599195