



## Case Report

# Successful Treatment of Red Ear Syndrome with Dual Therapy of Pregabalin and Duloxetine: A Case Report

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### Abstract

**Background:** Red ear syndrome is a rare disease, with nearly 100 cases reported worldwide. Red ear syndrome is generally refractory to treatment, and no definitive treatment plan has been established yet. **Case presentation:** In this case report, we present a 23-year-old male patient, known case of migraine, who was successfully treated with dual therapy of pregabalin and duloxetine for bilateral idiopathic red ear syndrome. **Conclusions:** To date, this is the first case presentation proving effective resolution of symptoms with dual treatment of pregabalin and duloxetine in bilateral idiopathic red ear syndrome. Further large-scale studies can potentially prove such outcomes.

**Keywords:** Red Ear Syndrome; Otalgia; Erythema; Pregabalin; Duloxetine

**Abbreviations:** RES: Red Ear Syndrome; MRI: Magnetic Resonance Imaging; Mg: Milligrams; TMJ: Temporomandibular Joint.

### Introduction

Red ear syndrome (RES) is a rare disease, with nearly 100 cases reported worldwide [1,2]. First described in 1994 by Lance, the pathophysiology remains unclear, although activation of the trigemino-autonomic circuit and dysfunction of cervical spinal nerves have been proposed [1-5].

RES typically presents as paroxysmal erythema of uni- or bilateral ear pinna, with or without burning sensation and otalgia, following triggers such as heat, touch, or neck movement [1-5]. RES is a clinical diagnosis, and diagnostic tests may include

magnetic resonance imaging (MRI) scan of the head and cervical spine, and orthodontic evaluation [1-3]. RES is generally refractory to treatment [1-5]. To date, this is the first case presentation proving effective resolution of symptoms with dual treatment of pregabalin and duloxetine in bilateral idiopathic red ear syndrome.

### Case Presentation

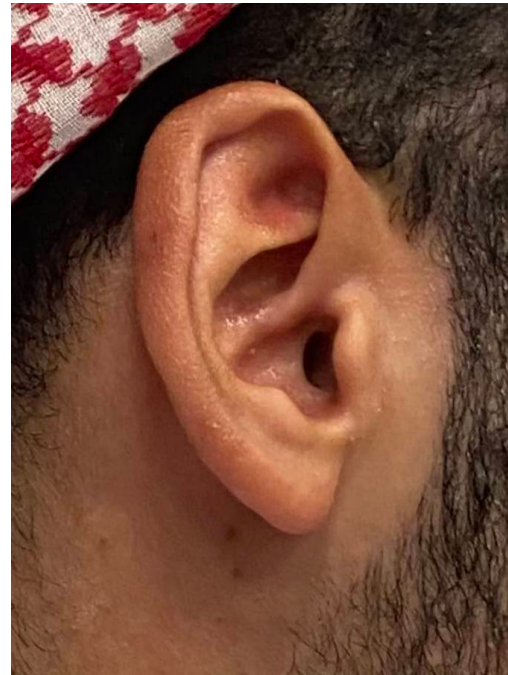
A 23-year-old male, known case of migraine, presented with complaints of chronic ear pain, burning sensation, and redness of both ears aggravated by cold air, head scarves, and side sleeping (Figure 1). He denied tinnitus or hearing loss. The patient was previously following with orthodontics for temporomandibular joint (TMJ) pain. Examination revealed erythema of bilateral ear pinna and mild tenderness. Otoloscopic examination was normal, and the oral cavity was clear. Magnetic resonance imaging showed normal temporomandibular joint morphology. The patient was cleared from rheumatology for autoimmune

diseases, from orthodontics as well as Ear Nose and Throat for TMJ abnormalities. From neurology clinic, the patient was given trials of amitriptyline, gabapentin, carbamazepine, and duloxetine monotherapy, with no benefit. Bilateral greater occipital, lesser occipital, auriculotemporal, supraorbital, and supratrochlear nerve blocks did not provide relief following a period of more than 8 weeks.



**Figure 1:** Erythema of external ear involving ear lobules, most pronounced in the helix, lobule and fossa triangularis.

Subsequently, dual therapy with pregabalin, 75 milligrams (mg) in the morning and 150 mg at night, plus duloxetine 60 mg once daily, were introduced along with lidocaine ointment. Within one week, there was resolution of all RES symptoms (Figure 2). Following that period, the patient attempted to take pregabalin and duloxetine as monotherapy, by himself, which caused the symptoms to reappear during a few days. Resumption of dual oral therapy suppressed symptoms the same week.



**Figure 2:** Resolution of ear erythema most notable in ear fossa triangularis and antihelix.

### Discussion

RES is classified into primary (idiopathic), and secondary RES which is structural [1-5]. Primary RES often accompanies migraine, as seen in our case, whilst secondary RES often accompanies TMJ dysfunction [1-5]. Our patient met RES diagnostic criteria, which were proposed by Lambru, et al [3]. RES was differentiated from relapsing polychondritis as episodes would last longer, ear lobule was not spared and there was no disfiguration of the cartilage [4]. Although RES is considered refractory to treatment [1-5], our patient demonstrated resolution of otalgia, burning sensation, and ear erythema during dual therapy with pregabalin and duloxetine, and RES symptoms were not brought on by light touch or neck movement.

Recent open-label trials suggested beneficial use of pregabalin, and other studies only noticed partial improvement [2, 3]. Conversely, gabapentin was mostly used demonstrating successful outcomes, and combination with verapamil recently showed complete resolution of RES symptoms [2]. Verapamil reduced frequency and severity of both RES and migraine episodes, whilst propranolol successfully treated RES secondary to cervical spondylosis, but not TMJD [2]. Few cases demonstrated complete response to non-steroidal anti-inflammatory drugs, such as indomethacin and ibuprofen, in both primary and secondary RES [4,5]. In patients with TMJ dysfunction, a dental plate was found to be curative [1,2,5]. Our patient's TMJ pain was controlled with a night guard.

Local nerve blockade and botulinum toxin type-a injections give partial relief [1-3]. Triptans, amitriptyline, and topical anesthetics were found ineffective or gave partial response [1-3, 5].

### Conclusion

Dual therapy with pregabalin and duloxetine could be a potential curative treatment for RES. However, large scale studies are needed to prove successful resolution of RES symptoms and specify the duration of therapy.

**Consent:** The patient has given their written informed consent for the case and images to be published.

**Clinical Message:** Dr. Nayla Al Khalifa, Dr. Manal Alsabbagh, and Dr. Mazen Raees aimed to demonstrate the effectiveness of treating RES with dual therapy of pregabalin and duloxetine.

**Author Contributions:** All authors have accepted responsibility for the entire content of this manuscript and approved its submission. Dr. Nayla Al Khalifa and Dr. Manal Al Sabbagh prepared the manuscript, whilst Dr. Mazen Raees supervised the patient's diagnosis, patient's management and manuscript.

**Author's Corner:** Dr. Nayla Al Khalifa, Dr. Manal Al Sabbagh, and Dr. Mazen Raees are satisfied with this research publication.

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**Data Availability Statement:** All data underlying the results are available as part of the article and no additional source data are required.

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