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Abstract

Eosinophilic otitis media (EOM) is characterized by a viscous middle ear effusion with eosinophilic predominance and tympanic perforation refractory to conventional management(1) which results in a substantially compromised quality of life because of either conductive or mixed hearing loss, chronic rhinosinusitis with nasal polyposis or eosinophilic asthma. (2)

The pathophysiological pathway begins with a Th2-type inflammation. Dupilumab is a humanized monoclonal antibody that is directed towards IL-4R α , thus decreasing the Th2 response by blocking the recognition of IL-4 and IL-13.

This case report will follow a patient with eosinophilic otitis media and a history of multiple surgical ear procedures. We present evidence of long-term follow up improvement in clinical, audiological, immunological, radiological and histopathologic characteristics and recommend the use of Dupilumab for the treatment of eosinophilic otitis media.

Keywords: Eosinophilic otitis media, eosinophils, Long-term follow-up, Dupilumab

Introduction

EOM is an entity characterized by recurrent and resistant otitis media. Lino, proposed the diagnostic criteria, which include a histological confirmation with eosinophil-dominant effusion. This pathologic entity is considered a type-2 (Th2) inflammatory disease, because it is characterized by an accumulation primarily of eosinophils in the middle ear mucosa associated with elevated levels of immunoglobulin E (Ig)-E, eosinophil cationic protein (ECP), interleukin (IL)-5 and eotaxin (5).

As a consequence of the inefficacy of traditional treatments and considering the pathophysiology of the disease, there has been a rising interest in the biologic management with therapy directed towards monoclonal antibodies.

Case presentation

We share the case of a 57-year-old male with a chief complaint of persistent otorrhea and progressive bilateral hearing loss. His past medical history was uncontrolled asthma, chronic sinusitis and nasal polyps with a severe Snot-22 score. He had had multiple surgical interventions. Physical examination was abundant otorrhea, bilateral tympanic perforation and inner ear polyps. His audiometry documented mixed bilateral hypoacusis, a CT scan showed an occupied mastoid with a soft-tissue density material in the epitympanum without erosion of the ossicular chain in both ears. Additional studies included the histologic confirmation of a predominantly eosinophilic middle ear effusion, negative allergy tests, negative specific IgE tests with a total IgE count of 288, a total eosinophil count of 540.

Management began with a 6-month dupilumab therapy after which a revisional mastoidectomy of the left ear was performed. Four months after said procedure, a type I tympanoplasty with a cartilage graft was performed, without any complications. At one year follow-up there was significant improvement in his clinical condition, his COMQ-12 (Chronic otitis media outcome test) score improved from 45pts to 15pts, he had a mild SNOT-22 score, closed bone air-gap, his audiometry had improved, he showed improvement in mastoid occupation as evidenced by a control CT and endoscopic evaluation.

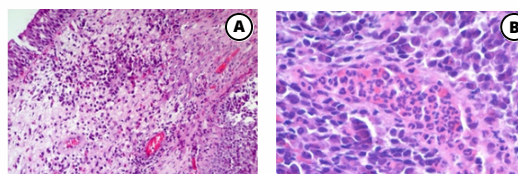


Figure 1. Hematoxylin & Eosin stain of the middle ear mucosa Pre and Post treatment b. (A) image shows 40 eosinophils per high-power field, after treatment (B) image shows 5 eosinophils per high-power field.

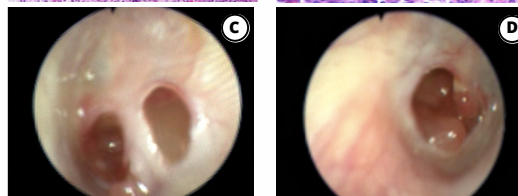


Figure 2. Otoendoscopy before treatment. Right ear shows polyps in the middle ear (C). Left ear shows tympanic perforation and edematous middle ear mucosa (D).

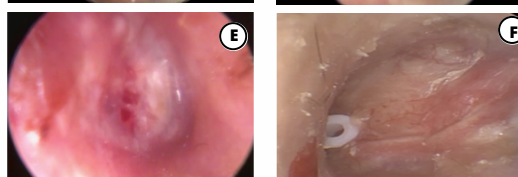


Figure 3. Otoendoscopy. Right ear shows an adequate closing of the tympanic perforation one-month post-op (E). The left ear shows an adequate integration of grafts without retraction, a correctly positioned ventilation tube. (F).

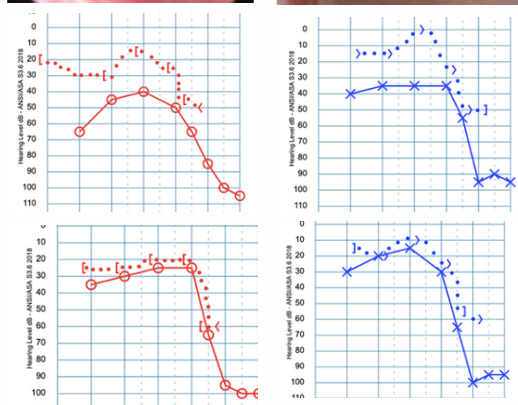


Figure 4. Preoperative audiometry shows moderate bilateral mixed hypoacusis.

Figure 5. Audiometry 3 months after the last surgical intervention and after a year of Dupilumab treatment. Shows complete gap closing and improvement in the PTA (pure tone average).

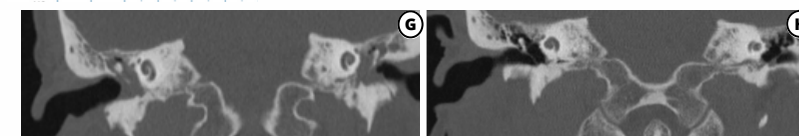


Figure 6 Coronal Ear CT Scan(G) previous (H) and 6 months after treatment, (G) showing bilateral soft tissue density occupation, low pneumatization of the mastoid, (H)Observes an adequate pneumatization of both middle ears and the described postoperative findings.

Discussion

Lino et al. published a series of 21 patients with chronic eosinophilic otitis media that relapsed after surgery (6). There has been a recent increase in interest in the development of specific immunotherapy, nonetheless, there has been no case reports published about joint management with Dupilumab, a humanized monoclonal antibody that acts upon 4R α lowering the Th2 response and revisional mastoidectomy, tympanoplasty and tympanostomy of the contralateral ear. The result is a significant improvement in audiological studies, notable changes in CT scan and endoscopy in relation to the disappearance of middle ear polyps, nose and paranasal sinuses.

Conclusion

EOM is a difficult to treat otitis media, our findings suggest a possible improvement in clinical outcomes in patients that present eosinophilic otitis media by combining immunotherapy, specifically Dupilumab and surgical management.

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