

En conclusión, queremos poner de manifiesto con esta serie de casos el importante papel que juegan los fármacos inmunosupresores en la producción de infecciones oportunistas graves, estando especialmente relacionada esta entidad con el uso de corticoides, anti-TNF o cuando se asocian varios, debiendo tenerla en cuenta ante la presencia de fiebre de origen desconocido en paciente con EI. No existe a pesar de ello gran evidencia científica, que relacione la infección por *L. monocytogenes* con la EI en tratamiento inmunosupresor no biológico.

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Ramsay-Hunt Syndrome in a patient with Crohn's disease under adalimumab: First case report



Síndrome de Ramsay-Hunt en un paciente con enfermedad de Crohn con tratamiento de adalimumab: Observación clínica

A 40 years old man with Crohn's disease (Montreal classification A2L3B3p), under azathioprine since 2004, was operated in 2007 for a complex perianal fistula and then started infliximab. Due to inefficacy of this therapy, he started adalimumab in 2008 with good response, but for personal reasons he withhold treatment in 2010. He needed another perianal surgery in February 2012 and treatment with adalimumab was resumed in March 2012. Four months later, he was referred because of persistent right otalgia and rubor of the auricular pavilion started a few days ago. Antibiotics were prescribed and he was discharged with the diagnosis of perichondritis. However, four days later he recurred to the Emergency Department due to clinical deterioration, with fever, vertigo, worsening of right otalgia, vesicular rash in auricular

pavilion, and ipsilateral face paresis (House-Brackmann grade IV). These new symptoms adding to the inefficacy of antibiotics were suggestive of a viral infection, and therefore he was admitted with the diagnosis of Ramsay-Hunt Syndrome. Despite our patient was under adalimumab, he was treated similarly to general population with steroids and acyclovir, and along with physiotherapy, he fully recovered from face paresis and cutaneous lesions.

Ramsay-Hunt Syndrome is a rare complication of latent Varicella-Zoster Virus (VZV) infection. Its pathophysiological mechanism is the infection of the geniculate ganglion of seventh cranial nerve by VZV, resulting in vesicular rash of the ear or mouth, facial paresis, vertigo, hearing loss and otalgia; sometimes, other cranial neuropathies may be present.

Inflammatory Bowel Disease (IBD) is a well-known risk factor for opportunistic infections.¹ Regarding herpes zoster, it was respectively the first and the second most frequent opportunistic infection in a cohort of IBD patients followed at Mayo Clinic² and in a multicenter Japanese cohort.³ Its overall incidence was 734/100,000 person-years in a large study of more than 100,000 patients with IBD.⁴ However, there are no reported cases of Ramsey-Hunt Syndrome in IBD patients.

Furthermore, this is also the first reported case of this complication of VZV reactivation in the setting of anti-TNF- α therapy. In one study, biological therapy was related to a higher risk of herpes zoster (OR 1.81, 95% Confidence Interval 1.48–2.21),⁴ but a recent meta-analysis failed to show elevated risk of herpes zoster in patients with Crohn's disease under biologics.⁵ Perhaps these drugs may contribute to more severe presentations of VZV reactivation, but the major concern in daily practice is probably related to the risk of appearance of a primary VZV infection in patients under these drugs, which can be fatal.⁶

ECCO guidelines¹ stated that non-vaccinated patients without a clear history of chickenpox or shingles must be tested for VZV-IgG, and that seronegative patients should be vaccinated at least three weeks before starting immunomodulators. Furthermore, since herpes zoster and infection by other viruses (as Herpes Simplex Viruses) are relatively common in IBD patients and knowing the potentially poor outcome in those under anti-TNF agents, it is crucial to have close monitoring. This can be achieved with an easy access to the Outpatient Clinic and perhaps with the telephone number of the clinician to allow early evaluation and guidance.

Patients with Ramsay-Hunt Syndrome have an overall poorer recovery when comparing to Bell palsy, and risk factors for a bad outcome include older age, diabetes, delayed treatment and a House-Brackmann grade more than IV. Our case illustrates that prompt diagnosis and therapy may promote a good outcome of this rare condition, even in patients under immunomodulators.

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