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LETTERS TO THE EDITOR

Evolution of a patient with submaxillitis secondary to azathioprine, 4 years later

Evolución de paciente con submaxilitis secundaria a azatioprina, 4 años después

Dear Editor,

We read with interest an opinion piece by Dr Blasco Patiño et al. recently published in GASTROENTEROLOGÍA Y HEPATOLOGÍA under the title ''Acute submandibular sialadenitis in a patient with Crohn's disease being treated with infliximab''.[1]

The authors reported the case of a patient with penetrating ileal impairment meeting the criteria for treatment with tumour necrosis factor (TNF) inhibitors. This patient started treatment with infliximab and, six weeks later, developed bilateral acute submaxillitis, which followed a good course after steroids were started. Their piece referred to a letter previously published in the same journal in which we reported the same complication in a patient with Crohn's disease of the ileum and colon, but in our case after starting treatment with azathioprine.

This complication relapsed after the drug was suspended and subsequently switched to mercaptopurine. Therefore, according to the score on the Naranjo scale, we determined that thiopurines were likely the origin of the patient's submaxillitis, possibly due to an idiosyncratic, dose-independent mechanism.

We agree with the authors on the difficulty of determining a direct pathophysiological mechanism accounting for the development of this condition. However, we disagree with Blasco et al. that the most plausible hypothesis was reactivation of an indeterminate virus with a special tropism for the salivary glands due to pharmacological immunosuppression (in their case due to infliximab and in ours due to azathioprine). For this reason, we would like to report the course of our case since November 2017, when it was published, following suspension of azathioprine/mercaptopurine. The patient, despite maintaining immunosuppressant treatment with TNF inhibitors, has not presented any further episodes of clinical submaxillitis. At present, he remains on maintenance biologic therapy, without having required any dose escalation, in clinical remission with mucosal healing demonstrated on his most recent colonoscopy, a year ago.

This course resembles that reported by Alves da Silva et al., who recently published another case of submaxillitis possibly linked to azathioprine, also resolved after switching to adalimumab.[2]

Finally, perhaps we should reflect on the origin of submaxillitis in patients with inflammatory bowel disease and even raise another possibility in addition to those discussed above (adverse drug effect versus immunosuppression), which would be that of an extraintestinal manifestation not reported to date in relation to disease activity; however, that would require a larger number of available cases for purposes of measuring statistically significant associations that account for the onset of this condition in these patients.

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References

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