LETTER TO THE EDITOR

Recurrent orbital myositis as an extra-intestinal manifestation of Crohn’s disease

Dear Sir,

Extra-intestinal manifestations (EIM) of Crohn’s disease (CD) affect up to 35% of patients and the presence of one EIM appears to be a predisposing factor for others. Orbital myositis (OM) is a subgroup of idiopathic orbital inflammatory syndrome involving one or more extra-ocular muscles and is described as a very rare EIM of CD.2

We present the case of a 55-year-old woman, with Crohn’s colitis since 1995, stricturing behavior, with type 2 arthropathy. She underwent a sigmoidectomy in 2004 for a 7-cm stenosis (histology of the surgical piece compatible with Crohn’s stenosis). She was treated with sulfasalazine and had no history of steroid therapy or other immunosuppressant therapy. Recurrent joint symptoms were difficult to control.

In 2008, while CD was in remission, she presented with severe right periorbital pain, worsening with eye movement; vertical diplopia with supraversion and infraversion impairment, no change in visual acuity, right periorbital edema, lid ptosis and exophthalmia. Pupils were equal and reactive to light. An orbital MRI revealed thickening and edema of the superior rectus muscle and levator palpebrae superioris muscle, with hypersignal in T2 and gadolinium contrast enhancement compatible with OM (Fig. 1). The patient was started on prednisolone 125 mg/day, reduced to 60 mg/day after 4 days, with rapid clinical improvement. Investigation ruled out other causes of OM. Two months later, while tapering steroid to 10 mg/day, she had a recurrent myositis affecting medial rectus of the left orbit, requiring an increase in the prednisolone dose (60 mg/day), with rapid clinical response. The steroid dose was then slowly tapered. Due to steroid-dependence, the patient was started on azathioprine, which was discontinued 3 weeks later due to severe gastrointestinal intolerance.

Escalation to anti-TNF biologic therapy was performed and she has been on infliximab for the last 2 years (induction regimen followed by a maintenance dose of 5 mg/kg every 8 weeks), with continued absence of ocular, joint and intestinal symptoms and no need for steroid therapy.

OM is an acute or recurrent inflammation of one or more extra-ocular muscles. A review of the few cases of Crohn’s-related OM described in the literature found a predominance of women, with CD of the colon or ileocolonic, which can be associated with other EIMs.3 The diagnosis is based on clinical findings in combination with characteristic changes on contrast MRI. High dose steroid therapy is the first-line treatment and typically leads to rapid resolution.4 In recurrent OM with steroid-dependence, infliximab (an anti-TNF monoclonal antibody approved for the treatment of CD) has been used with good results as steroid-sparing therapy, some of these cases being CD-related OM.3

Conflict of interest statement

The authors certify that they have no conflicts of interest to declare.

References


Rita Pimentel* Paula Lago Isabel Pedroto

Division of Gastroenterology, Centro Hospitalar do Porto, Oporto, Portugal

* Corresponding author.

E-mail address: pimentelrita80@gmail.com.

18 May 2012