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CORRESPONDENCE

Bilateral acute idiopathic panmyositis

Shirin Hamed Azzam[1] · Jimmy Uddin[1]

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To the Editor:

The most common presentation of orbital myositis is acute and unilateral, which initially responds to systemic corticosteroid therapy [1].

The aim of the current paper is to describe patients with bilateral idiopathic panmyositis who presented acutely as their first episode, which to our knowledge has not previously been reported.

Retrospective, descriptive case series of bilateral acute idiopathic panmyositis patients who were treated by the Adnexal department at Moorfields Eye Hospital between 2012 and 2019. Medical records of four consecutive patients met the clinical criteria. The case notes, imaging and response to treatment were analysed.

Four patients were diagnosed as having bilateral acute idiopathic panmyositis. Mean average was 47 years old (range 30–68 years old). Male to female ratio was 1:3. All patients had few weeks history of bilateral periorbital swelling and severe pain. Their examinations revealed marked chemosis with severe extraocular muscles (EOM) restriction in all gazes.

All our patients had laboratory investigations that included: Full blood count, Erythrocyte sedimentation rate, C-reactive protein, glucose, thyroid function test, thyroid antibodies, autoantibodies (ANA, ENA, NDna, ANCA), IgG4, and ACE levels that were normal. None of them had a history of thyroid eye disease or trauma and there were no signs of neoplasm, infection, collagen disease or Myasthenia gravis. Computed tomography (CT) scans showed enlargement of all EOM with tendons involvement (Figs. 1 and 2). All patients had brisk response after starting steroids treatment systemically. Three patients had a recurrence while tapering down below 30 mg daily. In these cases, biopsy was not performed as the diagnosis was made on clinical and radiological classic findings of bilateral panmyositis. They were managed successfully with methotrexate with no further flare up. Follow-up ranged from 5 to 24 months (mean, 12 months).

The classic presentation of myositis is unilateral painful ophthalmoplegia associated with signs of ocular inflammation and associated radiological findings. The symptoms and findings in our series are similar to previous reports regarding myositis [2, 3]. Although young adults in the third decade are most commonly affected in classic orbital myositis [1], in our series, bilateral panmyositis patients were older.

Previous reports showed that orbital myositis can recur on a different EOM on the contralateral orbit [4, 5]. In our series, all four patients presented acutely with bilateral symmetrical panmyositis, which has not been reported previously.

Based on our experience of bilateral acute panmyositis, we suggest high dose steroids initially, to start with 2–3 doses of intravenous methylprednisolone 500 mg (then oral prednisolone) or oral prednisolone (60 mg orally in a tapering regimen for few weeks depending on clinical response). Three of our patients had relapse, while tapering the steroids below 30 mg/day and had to be referred for second line immunosuppression treatment.

In conclusion, although the limitations of our study (which include a small sample due to the rarity of the disease and the lack of a control group), bilateral panmyositis, is a rare entity and can be made on clinical and radiological findings. Our cases responded more slowly to adequate dose of steroids than mono-myositis, and recurred in three quarters of the cases on reduction of steroids, which required 2nd line immunosuppression. Our investigations did not yield any underlying associated pathology.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Informed consent The authors confirm that written consent was obtained for the use of Fig. 2 in published media.

✉ Jimmy Uddin
jmuddin@aol.com

[1] Moorfields Eye Hospital, London, UK

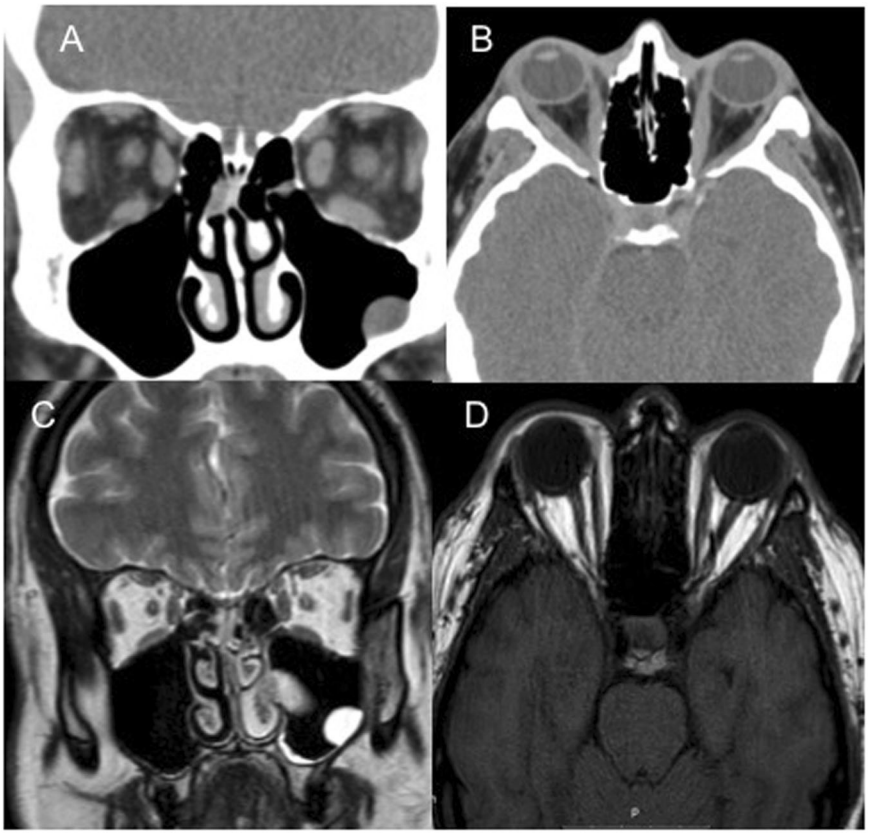

Fig. 1 Radiologic findings of bilateral panmyositis pre and post-treatment. a, **b** CT scan of a 32-year-old man that shows enlargement of all Extraocular muscles (EOM), including superior oblique and involving the tendons at presentation. **c**, **d** T1-MRI scan shows reduction in EOMs diameter post steroid treatment.

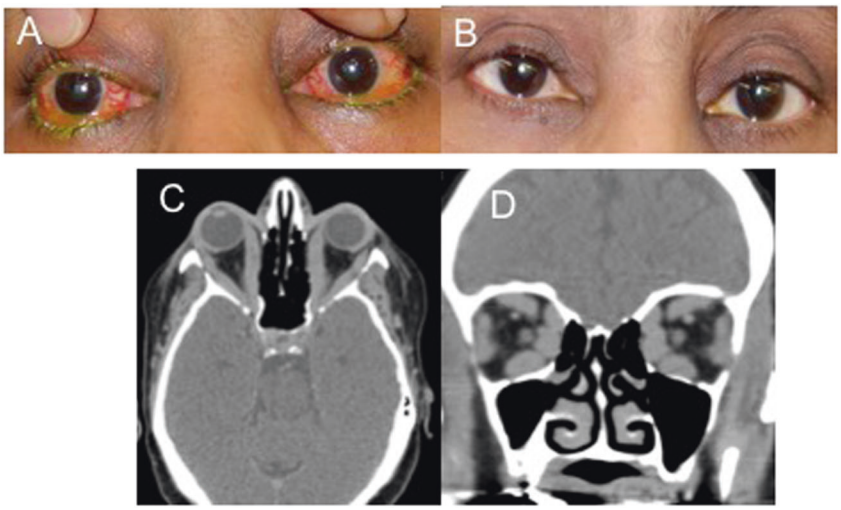

Fig. 2 Clinical findings of bilateral panmyositis pre and post-treatment. a A 60-year-old woman with bilateral dilated vessels at presentation. **b** Improvement post 2 weeks of oral Steroid treatment. **c**, **d** axial and coronal CT scans respectively, that shows enlargement of all Extraocular muscles (EOM) including obliques and involving the tendons. There is slightly dilated left superior ophthalmic vein. Patient was referred to Ultrasound that showed no reversal flow or enlargement of SOV.

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