Isolated lateral rectus myositis

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ABSTRACT

يعد التهاب عضلات الحجاج من الالتهابات النادرة الغير حبيبية التي تحدث في حجاج العين، ويعد كلاً من مرض غريف والاضطرابات التكاثرية اللمفاوية من أكثر مسببات المرض شيوعاً، ولذلك يجب معرفة أسباب المرض وأخذها بعين الاعتبار وذلك بعد استثناء الأسباب المعروفة لهذا المرض. يعد الالتهاب المعزول لعضلات الحجاج (Isolated orbital myositis) من الأشكال النادرة لهذا المرض. نستعرض في هذا المقال حالة مريضة مصابة بالتهاب معزول للعضلة المستقيمة في الحجاج من أجل لفت انتباه الأطباء إلى هذه الحالة حيث أن المبادرة بالعلاج أدى إلى شفائها بالكامل.

Orbital myositis is a rare non-granulomatous inflammatory process within the orbit. Grave's disease and lymphoproliferative disorders are considered the most common cause of orbital myositis. The idiopathic form should be considered after exclusion of known causes or associations. Isolated orbital myositis is a very rare form of this disease. We report a case of an isolated lateral rectus myositis to draw the attention of physicians to this condition, as prompt treatment in our patient resulted in complete recovery.

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Idiopathic orbital inflammation is a non-infectious, non-neoplastic, space occupying inflammatory lesion without identifiable local or systemic cause. The condition is called orbital myositis when it involves the extraocular muscles.[1] Isolated orbital myositis (IOM) is a rare disease, which affects mainly females. Secondary causes should be excluded before reaching this diagnosis.[1] A high index of suspicion is required for proper management. The following case is discussed to highlight the clinical presentation and management.

Case Report. A 27-year-old Saudi healthy female presented to the Emergency Department of King Khalid National Guard Hospital, Jeddah, Saudi Arabia complaining of left sided temporal headache for 3 days. The pain progressed from a mild dull ache to severe pain, which increased with eye movement. She noted lateral limitation of left eye movement, which was associated with diplopia. She had no fever, photophobia, history of migraine, or trauma, and no symptoms suggestive of any underlying systemic disease. No history of previous upper respiratory tract infection, oral or genital ulcers, or similar illness in the past. Physical examination revealed a healthy woman with normal vital signs. She was conscious with normal higher cortical functions. She had no goiter or lymphadenopathy. Eye examination revealed no chemosis, conjunctival injection, ptosis, or proptosis of either eyes. The pupils were equal and reactive. Left eye abduction was painful and limited with diplopia (**Figure 1a**). Funduscopic examination showed a normal disc. The rest of the physical examination was normal. Investigations showed normal complete blood count, erythrocyte sedimentation rate (ESR) was 50mm/hr (normal: 0-15mm/hr), and C-reactive protein (CRP) was 6.2 mg/L (normal: 0-5mg/L). Her biochemical profile was normal. Antinuclear antibody was 0.2 (<1 index), endomysial antibody was 2.7 (<25 u/ml), complement 4 was 0.23 (0.1-0.4 g/l), and complement 3 was 1.11 (0.9-1.8 g/l). Rheumatoid factor level, thyroglobulin antibodies, anti thyroperoxidase antibodies, and angiotensin converting enzyme levels were negative. In addition, HIV and herpes serology were negative. The CSF composition and pressure were normal. Tensilon test was negative, and thyroid function tests were normal. Her chest x-ray was unrevealing. The CT and MRI of brain were normal. An MRI of the orbit showed thickening of the lateral rectus muscle with subtle enhancement (**Figure 2**). She was diagnosed as an isolated left lateral rectus myositis after ruling out common causes. She was treated by prednisolone 60 mg

Figure 1 - The patient looking to the left with **A)** limitation of abduction in the left eye. **B)** Full recovery after treatment.

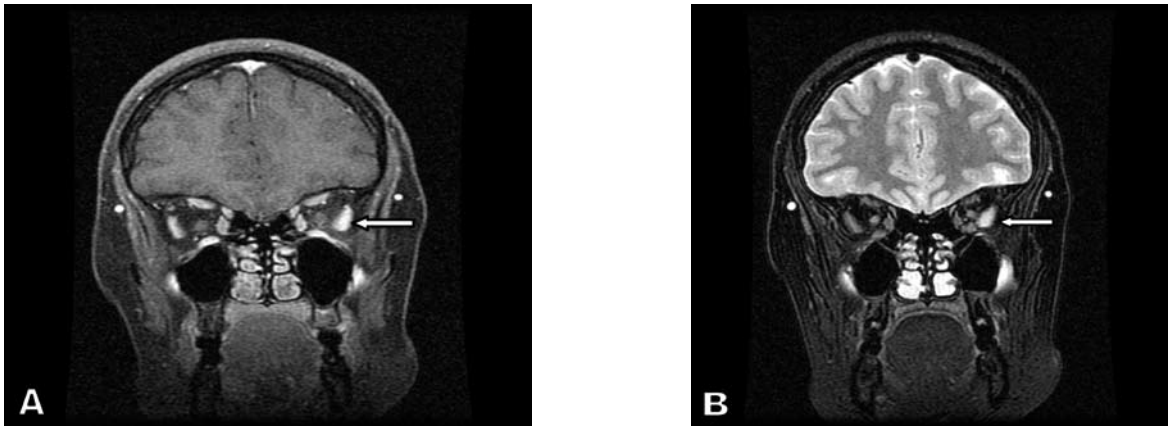

Figure 2 - Patient MRI showing **A)** post gadolinium contrast with fat suppression T1, and **B)** Coronal images in T2, demonstrating thickened left lateral rectus muscle (arrows), increased signal intensity in T2 and abnormal enhancement.

daily, which was started the day after her presentation. Prednisolone was then tapered gradually over 3 months. Her symptoms improved, and the ESR and CRP normalized. Follow up MRI of the orbital muscle was normal. During subsequent follow up in the clinic over 12 months, she was asymptomatic with normal function of the extraocular muscles (**Figure 1b**).

Discussion. Idiopathic orbital inflammation (pseudotumor) is a non-granulomatous, inflammatory process within the orbit that causes a variable degree of polymorphous infiltrate and fibrosis involving the orbit diffusely or targeting a specific anatomic structure.[1] Orbital myositis is a subgroup of this broad clinical category in the absence of thyroid disease. Idiopathic orbital inflammation was initially described in 1903 by Gleason.[1] It is mainly a disease of females. It may be acute, sub acute, or chronic, and a recurrent tendency has been reported even with treatment.[2,3]

Secondary causes of IOM such as Graves's disease, lymphoproliferative disease, as well as other conditions, which include connective tissue disease (systemic lupus erythematosus), ulcerative disease, infections such as herpes zoster and paraneoplastic syndrome, should be excluded.[4-7] In our case no secondary causes were found, and follow up over one year did not reveal any underling disease.

In the emergency department, CT is the modality of choice for diagnosing orbital myositis. This is characterized by enlargement of the extraocular muscles extending interiorly to involve the tendon insertion on the globe. However, absence of tendon involvement does not rule out idiopathic orbital myositis.[8] An MRI is considered as an alternative diagnostic tool.[9] The MRI signs of orbital myositis are more obvious and sensitive than CT as illustrated in our case.

Corticosteroid is the mainstay of treatment, although recurrent attacks were reported.[2,3] Radiation therapy has been employed in refractory cases to corticosteroid, however, its rational for improving symptoms, and its efficacy is unclear.[10] Keeping in mind that if the patient presents with recurrence, it is important to rule out

secondary causes again before considering any second line of treatment. Chemotherapy, radiation, botox, and surgery are other modalities for treatment.[1,2] In some reports, nonsteroidal anti-inflammatory drugs can be used effectively as first line.[10] We used corticosteroids as the treatment of choice for our patient, which showed quick and excellent response without recurrence for one year of follow up.

Our purpose in reporting this case is to draw the attention of physicians to this rare entity, which may cause a diagnostic dilemma. Recognition of this condition with prompt treatment will yield an impressive response in most cases, as illustrated in ours.

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Case Reports

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