

Case Report*Open Access, Volume 6***Rare presentation of periscleritis as painful ophthalmoplegia:
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Introduction

A middle-aged diabetic female presented with acute headache, left eye ptosis, pain and ophthalmoplegia. The best corrected visual acuity in right eye was 20/20 and left eye was 20/200. Anterior segment evaluation was unremarkable. Extraocular movements in the left eye were restricted in all the gazes except levoduction. Ultrasonography showed retinal choroidal thickening and the retrobulbar echo lucent area. MRI brain showed ring sign in both eyes. Infectious and autoimmune causes were ruled out. Patient was diagnosed with idiopathic bilateral periscleritis with associated third nerve palsy in the left eye and started on pulse dose of methylprednisolone followed by oral steroids in tapering dose and oral azathioprine. The patient showed a rapid recovery within three days. This case highlights a rare presentation of Idiopathic Orbital Inflammation (IOI), emphasizes the role of orbital imaging in diagnosis and importance of timely immunomodulatory therapy.

Background

Idiopathic Orbital Inflammation (IOI) accounts for 4.7% to 6.3% of orbital diseases is a benign, non-infectious, non-neoplastic inflammatory disorder involving orbital tissues. While

myositis is the most common localized form, involvement of Tenon's capsule, sclera or peri scleral tissues (periscleritis) is rare [1]. Painful ophthalmoplegia is a diagnostic challenge and requires prompt differentiation from life threatening causes such as cavernous sinus thrombosis, vasculitis, neoplasia [2]. IOI presenting as painful ophthalmoplegia and localised periscleritis has not been reported earlier and hence deserves a mention. IOI is a diagnosis of exclusion and a high index of suspicion is required to diagnose the localised form of IOI. The patient was given systemic immunosuppressive therapy, after ruling out the infective causes, to which the patient responded remarkably.

Case presentation

A middle-aged women with well controlled type 2 diabetes mellitus presented to the medical emergency department with headache, and left sided severe retro orbital pain, drooping of the left eyelid and restricted eye movements for eight days. There was no history of trauma, fever or any migraine preceding the illness. On examination, her visual acuity was 20/20 in OD and 20/200 in OS. Pupillary reflexes were intact bilaterally. The left eye had complete ptosis and limitation of all extraocular movements except levoduction consistent with pupil-sparing third nerve palsy (Figure 1). The left eye had fair levator palpe-

bral superioris action of 6 mm. The left eye examination showed 1+ vitreous cells (SUN classification). Her anterior segment was remarkable in both eyes. A provisional diagnosis of left eye pupil-sparing painful ophthalmoplegia with vitritis was made.

Investigations

The ultrasonography of both eyes revealed bilateral retinal-choroidal thickening with retrobulbar echo-lucent area consistent with the 'T-sign' and point-like echoes in the posterior vitreous in the left eye (Figure 2). Systemic investigations revealed raised erythrocyte sedimentation rate. Complete blood counts, thyroid profile, blood cultures, viral serology, HLA B-27 and Treponema pallidum haemagglutination test and fasting blood sugar levels were unremarkable. Autoimmune screening was negative for ANA, ANCA, ACE and RA factor. MRI head revealed hyperintense retrobulbar fat suggestive of inflammation. Another retrobulbar hypointense area in both eyes was consistent with a "ring sign" suggestive of bilateral periscleritis (Figure 3). There was no evidence of any intracranial aneurysm or tumour. The cavernous sinus was normal.

Differential diagnosis

The common causes of painful ophthalmoplegia include neoplasms (primary intracranial tumours or distant metastasis), and vascular causes (aneurysms, carotid dissection, carotid-cavernous fistula). Radiological investigations clinched the diagnosis of IOI in view of the localised idiopathic orbital inflammation presenting as bilateral periscleritis. Other associations of IOI like Crohn's disease, Streptococcal or viral infection, psoriasis and ankylosing spondylitis were excluded.

Outcome and follow-up

The patient showed a rapid recovery within three days. There was an improvement in vision, pain, ptosis, and eye movements (Figure 4). The immunosuppressive therapy was continued for 12 months. The patient has not shown recurrence of the IOI at 2-year follow-up.



Figure 1: The left eye had complete ptosis and limitation of all extraocular movements except levoduction.

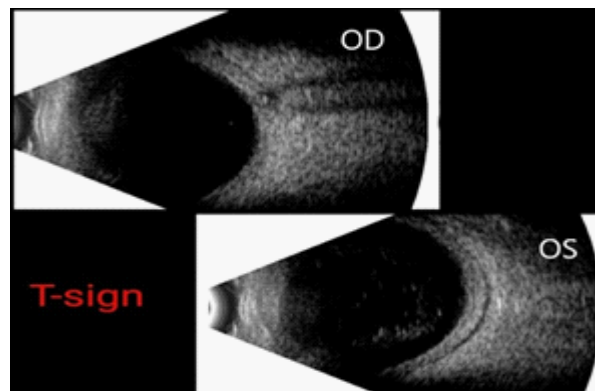


Figure 2: Bilateral retinal-choroidal thickening with retrobulbar echo-lucent area consistent with the 'T-sign' and point-like echoes in the posterior vitreous in the left eye.

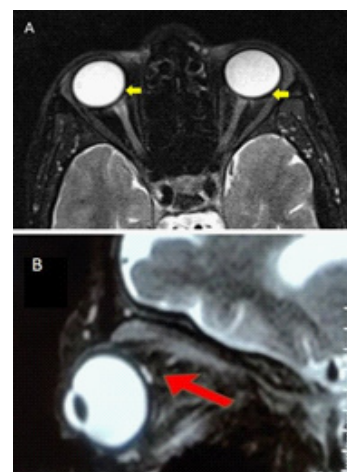


Figure 3: Retrobulbar hypointense area in both eyes was consistent with a "ring sign" suggestive of bilateral periscleritis.



Figure 4: Improvement in eye movements.

Discussion

Idiopathic orbital inflammation has a varied presentation. Apart from diffuse orbital involvement which presents as proptosis, the localised form of the disease includes isolated myositis, dacryoadenitis, scleritis, periscleritis, uveitis or focal mass. Our case presented with painful ophthalmoplegia and the radiological investigations clinched the diagnosis of IOI. The usual causes of painful ophthalmoplegia include neoplastic causes (intracranial primary neoplasm or secondaries in the brain), or

vascular causes in the brain (aneurysm, CCF, carotid dissection) which warrant an urgent MRI. In the index case, the radiological investigations clinched the diagnosis of bilateral peri-scleritis. The painful ophthalmoplegia could be due to the inflammatory changes in the tight connective tissue of the orbit could exert pressure on the third and fifth nerves in the orbit leading to painful ophthalmoplegia. Bilaterality is seen in 30% of cases of IOI [3]. In the case of IOI, one must exclude infections like Streptococcal respiratory tract infections, Lymes disease, viral infections like varicella zoster and other systemic inflammatory disease causing posterior scleritis. Systemic steroids with slow taper have been the established first-line treatment [3]. Rapid response to treatment is known in 60% of the cases but 20% recur and need an additional immunosuppressive agent. Azathioprine is effective as a steroid sparing agent in IOI, especially in recurrent or corticosteroid dependent cases [4]. Early immunosuppression in our patient enabled sustained remission without recurrence.

Learning points/Take home messages

1. Localised periscleritis in Idiopathic orbital inflammation can present as painful ophthalmoplegia.
2. Imaging is diagnostic for localised periscleritis in cases of IOI.
3. Infectious and autoimmune causes must be excluded before initiating steroids.
4. Early diagnosis and immunosuppression can give gratifying results.

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