IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 22, Issue 1 Ser.11 (January. 2023), PP 38-40
www.iosrjournals.org

Nonspecific Orbital Inflammation (NSOI) – A Rare Presentation in Elderly

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ABSTRACT
Nonspecific orbital inflammation (NSOI) is a benign inflammatory process of the orbit. NSOI is characterized by polymorphous lymphoid infiltrate with varying degrees of fibrosis, without any local or systemic cause. It is a diagnosis of exclusion and commonly observed in middle aged individuals. We report a case of an 82-year-old diabetic male with NSOI. Treatment with systemic prednisolone showed complete resolution. 6 months follow-up has shown no recurrence. This case is being presented since NSOI in elderly is a rarity.
KEYWORDS: *Orbital pseudotumor, NSOI, orbital cellulitis, elderly*

Date of Submission: 12-01-2023 Date of Acceptance: 28-01-2023

I. Introduction

Nonspecific orbital inflammatory disease (NSOI) is as an idiopathic tumourlike inflammation with pleomorphic inflammatory cellular response and a fibrovascular tissue reaction. Since it mimics awide range of pathologic conditions,misdiagnosis and inappropriate treatment arenot infrequent. However, thepotential for permanent visualdysfunction makes orbital pseudotumor anophthalmologic process that must not be overlooked. We present a case of orbitalpseudotumor that occurred in an elderly, diabetic male mimicking orbital cellulitis.

II. Case Report

An 82-year-old male presented with complaints of swelling and redness around left eye (LE) for 1 month and pain for 2 days. He gavea history of painless, progressivedefective vision in LE for 6 months.He was a known case of Type 2 Diabetes mellitus for 5 years under insulin therapy, systemic hypertension for 2 years.There was no history of fever, rash, joint pain, weakness in the limbs, preceding infection or trauma to the eye. There was no history of use of any medications (bisphosphonates, lithium and chemotherapeutics). He denied any similar episodes in the past. General and systemic examination was normal. Blood pressure was 130/80mmHg recorded in sitting position in right upper arm. Right eye (RE) intraocular pressure was 15mmHg, best corrected visual acuity was 6/9 and examination was within normal limits. LE intraocular pressure was 16mmHg, visual acuitywas perception of light and accurate projection of rays.

Figure 1(a) Left eye (LE) revealing restricted dextroelevation (b) restricted dextro version (c) dextro depression restriction in LE.

Ocular examination of LE showed 30-degree exotropia, restricted eye movements superiorly, temporally and medially. [Figure 1] Lid was oedematous, severe ptosis of 6 mm, axial proptosis of 4 mm (21mm).Conjunctiva was congested and chemosed. Normal pupillary light reflex and mature lens was present. [Figure 2]A tender, non-palpable swelling noted in supra lateral orbital rim with no audible bruit. Ultrasound B scan revealed no posterior segment abnormalities.

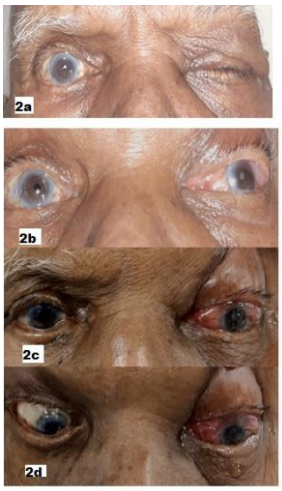

Figure 2 (a) LE reveals severe ptosis, lid oedema (b) restricted up gaze (c) primary gaze shows 30-degree exotropia, chemosed conjunctiva (d) restricted down gaze. RE within normal limits.
Figure 3 Table showing investigations of the patient.

An empirical diagnosis of orbital cellulitis was made and started on antibiotic therapy. Investigation revealed normal liver, renal and thyroid functions with significant eosinophilia and elevated erythrocyte sedimentation rate(ESR). [Figure 3]

INVESTIGATION	RESULT
Haemoglobin g/dl	7.4 g/dl
FBS mg/dl	90 mg/dl
PPBS mg/dl	142mg/dl
HBA1c %	7.9 %
Platelet /cu mm	1,55,000/cu mm
RBC/cu mm	2.56 million/cu mm
WBC cells/cu mm	4600 cells /cu mm
Eosinophils	9.8 %
ESR @ 30 mins	74
ESR @ 1 hour	150mm/hr
RFT – Renal Function Test	
Blood Urea	36 mg/dl
Serum Creatine	1.0 mg/dl
Liver Function Test	
ALT	27 IU / L
Total Bilirubin	0.3 mg
Thyroid Function Test	
TSH	2.6 m IU / L
Peripheral smear	**Normocytic normochromic anemia**
Blood culture	**Negative**

Figure 3 Table showing investigations of the patient.

Imaging showed normal chest X ray and ultrasound abdomen and KUB.T2 weighted Magnetic Resonance imaging (T2W MRI) revealed clear sinuses, intraconal and extraconal soft tissue inflammation, peri orbital soft tissue inflammatory changes, pre-septal and post septal inflammation and thickening,muscle tendon involvement with bulky lacrimal gland.[Figure 4]

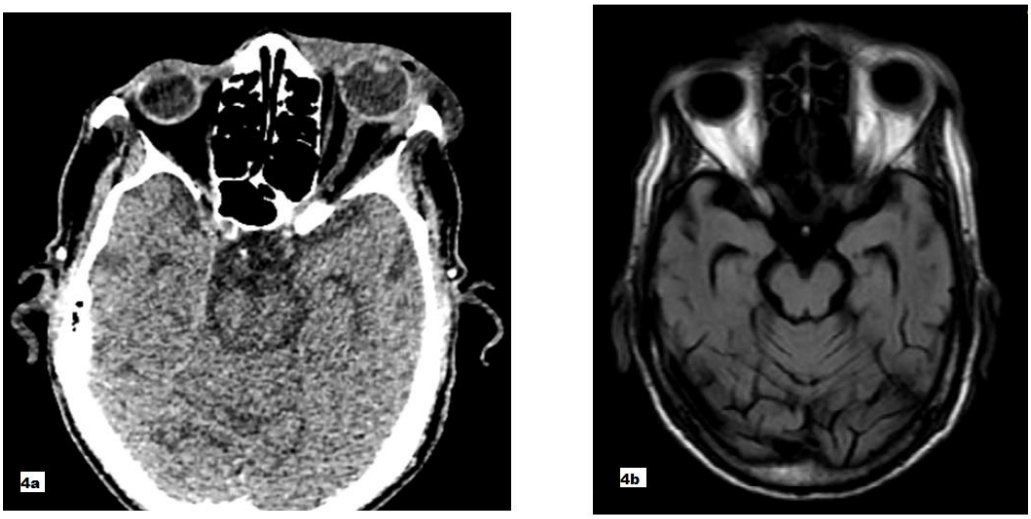

Figure 4 (a) Computed Tomography (CT) brain, orbit and paranasal sinuses - Axial section showing LE diffuse periorbital soft tissue thickening , bulky lacrimal gland, clear sinus cavity.
(b) Magnetic Resonance Imaging T2 weighted FLAIR (Fluid- Attenuated Inversion Recovery) sequence revealing white hypodense intraconal and extraconal soft tissue thickening .

With the above clinical scenario, elevated ESR,eosinophilia, clear sinuses with bulky lacrimal gland, muscle tendon involvement and no infective aetiologythe case was diagnosed as NSOI.

Patient was started on oral prednisolone (1 mg/kg/day) for 2 weeks followed by tapering doses over 10-12 weeks. Patient showed dramatic response within 48 h, ptosis improvement within 2 days and at the end of 1 week, improvement in eye movements. [Figure 5]

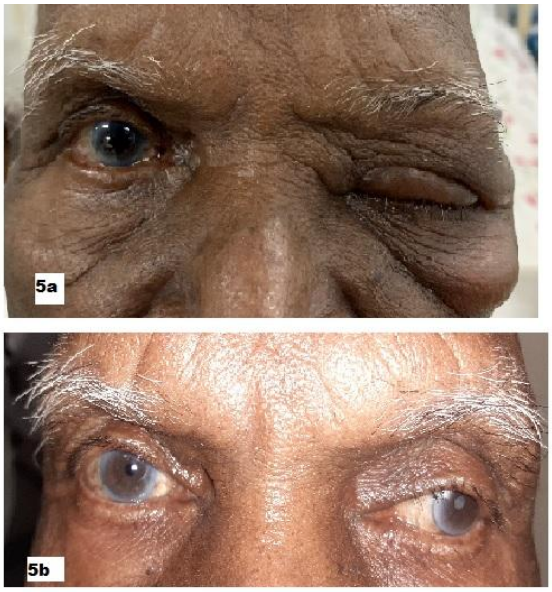

Figure 5 (a) Pre-steroid phase – clinical image revealing severe ptosis (b) 2 weeks post steroid therapy – reduction in chemosis, lid oedema

The patient was discharged on maintenance dose of steroidsand was kept under observation with monthly follow-up for recurrence or disease progression.One-year follow-up has shown no recurrence. This case stands unique as NSOI in elderly is a rarity.

III. Discussion

NSOI, an idiopathic non-infectious inflammation of orbit,is the third most common orbital disease following Graves ophthalmopathy and lymphoproliferative diseases. Described first by Brischfield in 1905,it is commonly observedin middle age.[1,2]

Aetiology of NSOI is still emerging.[1] It is alternately believed to be a subclinical infection or an immune process secondary to viral upper respiratory tract infection.[3,4]Though NSOI is primarily a diagnosis of exclusion, clinical features, lab investigations and imaging contribute to the diagnosis.[2]

Unilateral periorbital pain, cranial nerve palsies and a dramatic response to corticosteroids therapy are the hallmarks of clinical presentation in orbital pseudotumor. [1–3]Clinical features such as periorbital oedema, painful eye movements, erythema, proptosis, ptosis or diplopia understandably mimic orbital cellulitis, retrobulbar abscess or hematoma,Graves' disease, sarcoidosis, orbital tumours, Wegner granulomatosis and vasculitis.[1,4]

Inflammation of extraocular muscle can be acute, subacute or recurrent and commonly involve the medial rectus muscle.[1,3]The involvement of muscle tendon helps distinguish NSOI from thyroid orbitopathy.[4]There are no specific tests for NSOI but elevated erythrocyte sedimentation rate , eosinophilia are suggestive of inflammatory aetiology of NSOI. [1,3,5]

Imaging of the orbit is best done by CT-scan or MRI. Depending on the structures involved, imaging may reveal a diffuse or discrete orbital mass, uveoscleral thickening, contrast enhancement of Tenon's potential space, infiltration of adjacent fat, periocular tissues or optic nerve and extraocular muscle enlargement including the muscle tendon.[1–3,5]However, identifying the true nature of this condition and recognizing it from other malignant pathologies is imperative to evade needless invasive intervention.[3,5]

Systemic corticosteroids therapy (oral prednisolone acetate 1.0 to 2.0mg/kg/day) is the mainstay of treatment.Dramatic improvement following steroids has been recorded in over 70% of the cases.[2,3,5]There is no established protocol for steroid therapy but gradual tapering over weeks to months with monitoring of clinical features and glycemia is recommended.[1,3,5]In cases non responsive to steroids, methotrexate, cyclophosphamides or infliximab have been of some value.[5] Almost 1/3rd of those with NSOI may have persisting deficit with a small minority (2%) resistant to treatment. [1,3] The reported patient showed good response to steroids and no relapse at 1 year.

IV. Conclusion

NSOI being apotential vision-threatening disease, prompt and early management is crucial. A high index of suspicion and meticulous monitoring is essential to identify atypical presentation.

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