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Case Report

Unilateral total ophthalmoplegia with proptosis and loss of vision in a patient of herpes zoster ophthalmicus: A rare case report

Tulika Gupta, Shikhar Ganjoo*, Neeraj Sharma, Harvinder, Ashok Kumar Khurana

Department of Ophthalmology, Shree Guru Gobind Singh Tricentenary Medical College and Hospital, Gurugram, India.
* Department of Dermatology, Shree Guru Gobind Singh Tricentenary Medical College and Hospital, Gurugram, India.

Abstract Herpes zoster Ophthalmicus accounts for a minority of all patients with zoster infections. It leads to varied clinical presentations, but total unilateral ophthalmoplegia has rarely been reported in the literature. We hereby present a 50-year-old male patient presenting with the above combination for aiding the clinical diagnosis by dermatologists and ophthalmologists. Early initiation of treatment leads to a near total recovery of ophthalmoplegia in the majority of treated patients.

Key words
Unilateral total ophthalmoplegia, proptosis, herpes zoster ophthalmicus.

Introduction

Herpes zoster Ophthalmicus, also popularly known as Ophthalmic zoster, is a condition that primarily affects the ophthalmic division of the trigeminal nerve. It occurs due to the reactivation of the latent varicella-zoster virus involving the eyes or its surrounding area. Herpes Zoster Ophthalmicus (HZO) accounts for about 10-20% of cases of Varicella-zoster infection.[1] It leads to various clinical manifestations such as erythematous rash, which progresses to a maculopapular rash, vesicular rash, ptosis, occlusive vasculitis causing ischemic neuropathy and extraocular eye muscle palsies.[2] Occurrence of total unilateral ophthalmoplegia, with proptosis, loss of vision and impaired ocular ductions in all four directions is rarely reported.[3]

Address for correspondence
Dr. Shikhar Ganjoo, Associate Professor,
Department of Dermatology & STD,
Shree Guru Gobind Singh Tricentenary Medical College and Hospital, Gurugram, India.
Ph: +919891153924
Email: shikhar.ganjoo@gmail.com

Case report

A 50-year-old male patient presented to the eye outpatient department with sudden vision loss, drooping of the upper eyelid, inability to open the eye, restricted eye movement in all directions, mild pain, swelling of the left eye and cheek, protrusion of the left eyeball for the last two days. Two days later, few vesiculo-papular eruptions were seen on the tip of the nose respecting the midline, which became more erythematous and increased in number involving the upper lip on the left side **(Figure 1)**.

The patient reported a history of fever which started 20 days back. Seven days after the onset of fever, he tested positive for COVID-19 (by RT-PCR). The patient was ambulatory and isolated himself since he was febrile. Fever was not associated with any respiratory symptoms; therefore, he did not take any other treatment

Figure 1 Vesiculo-papular eruptions seen on the tip of the nose respecting the midline, drooping of the upper eyelid, inability to open the eye, swelling of the left eye and cheek and protrusion of the left eyeball.

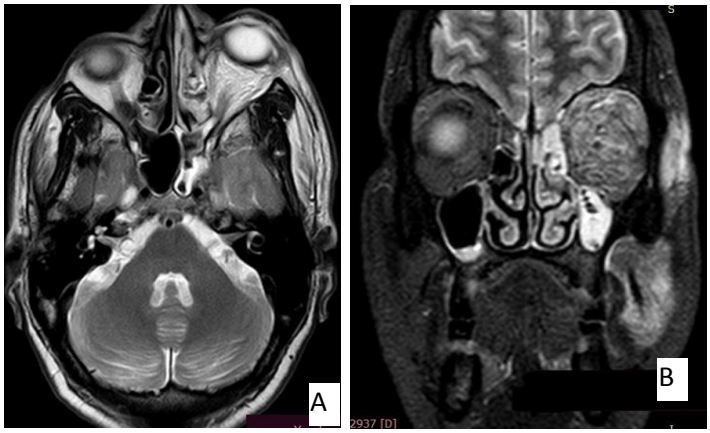

Figure 2 show STIR and T2 hyper intensity in left Maxillary Sinus and left Retro Orbital Region pushing the left eye ball anteriorly. A) T2 weighted Axial. B) (Short tau inversion recovery) STIR Coronal images.

Table 1 Ocular Examination of the patient.

	Right	*Left*
Visual acuity	6/60	PL negative
Position of eyeball	NAD	Mild axial proptosis
Adnexa	NAD	Erythema with Lid edema Upper lid > Lower lid Severe ptosis (Complete closure of the eye)
Extraocular muscles	NAD	Complete ophthalmoplegia (No movement present in any direction)
Conjunctiva	NAD	Superficial congestion with chemosis
Cornea	NAD	Normal size, shape, surface, and sheen
Anterior chamber	NAD	NAD
Pupil	NAD	Mid dilated, reaction to light absent (direct and consensual)
Lens	Early cataractous changes	Early cataractous changes
Vitreous	NAD	NAD
Fundus	Optic Disc- WNL CDR = 0. 4:1 FR + No other lesion seen	Optic disc-WNL CDR= 0.7:1 FR -dull No other lesion seen
Skin overlying the eye, nose, and sinus		Erythematous Papulovesicular lesions are present on the left side of the nose, tip of the nose, upper lip respecting the midline.

like steroids, oxygen, Remdesivir, or tocilizumab. No fever was observed five days before the presentation.

The patient presented with a history of Diabetes mellitus over the previous two years, an elevated blood sugar of 300 mg/dl, along with glycosuria and raised HbA1c (14.6%). CRP and ESR were also raised; they were 92 and 32, respectively. There was no history of hypertension or Tuberculosis. Hemogram turned out to be expected and Blood culture did not show any growth after five days. Diagnostic nasal endoscopy did not reveal any changes or any discharge. A nasal swab for (Potassium hydroxide) KOH and biopsy from the anterior surface of the middle turbinate was also negative. PL (perception of light) was absent on examination of the left eye. The patient had mild axial proptosis with eyelid edema, erythema, and

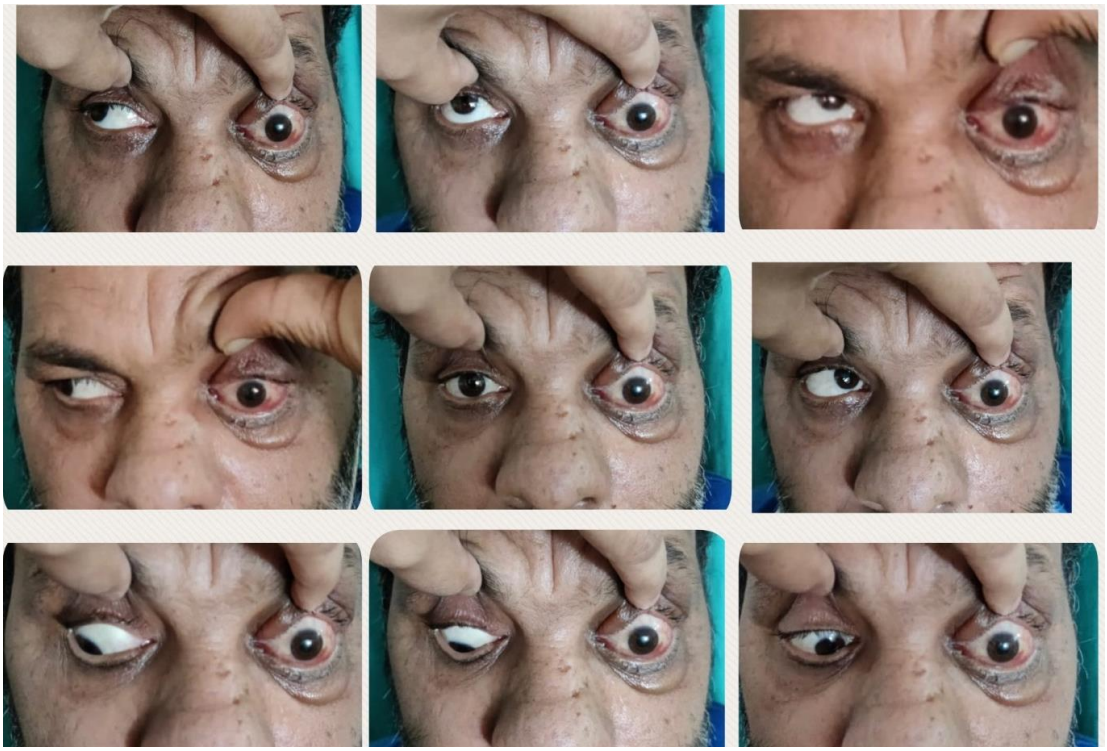

Figure 3 Figure showing 9 cardinal gazes pictures of the patient demonstrating that extraocular movements are absent in the left eye (involved eye).

severe ptosis, along with complete ophthalmoplegia. Conjunctival chemosis was present with superficial congestion. The pupil was mid dilated and reaction to light was absent. On fundus examination, the cup disc ratio (CDR) was 0.7:1, the foveal reflex was dull, but no other lesions were seen. The detailed examination of both eyes is tabulated below **(Table 1)**.

Contrast-enhanced computed tomography of paranasal sinuses (CECT PNS) with orbit showed evidence of inflammatory changes in the form of soft tissue shadows and fat stranding in the retrobulbar region in left orbit, pushing the globe anteriorly causing proptosis. Lateral and superior rectus muscles appear thickened with fuzzy edges. The soft tissue shadow showed mild and patchy heterogeneous enhancement. The optic nerve was unremarkable, and there was no bony erosion ruling out mucormycosis as the cause of orbital involvement **(Figure 2)**.

The patient was given intravenous Amoxicillin and Clavulanic acid (Augmentin), amikacin, and metronidazole empirically. Blood glucose monitoring was done every four hours, and the values were in the range of 383, 227, 452 mg/dl even after starting insulin. Dosages of regular and Lantus insulin were regulated according to the blood sugar levels.

A diagnosis of left orbital cellulitis with Herpes Zoster Ophthalmicus was made. The patient was initially prescribed Acyclovir 800 mg 5 times/ day. After two days, he was switched over to Valacyclovir 1g three times a day. The skin lesions markedly improved and the proptosis decreased substantially. The extraocular movement also started soon after the patient moved his eye superiorly and lifted his ptotic upper lid **(Figure 3)**.

Discussion

Varicella zoster virus VZV is a double-stranded DNA virus. Virus-laden respiratory droplets initially invade the upper respiratory tract and undergo replication, adjacent lymph nodes are invaded thereafter. Around seven days later, progression to secondary cutaneous infection is seen because of virus dissemination manifesting as a vesicular eruption. Through retrograde axonal transport, the virus enters the sensory nerve cell bodies in ganglia during primary infection, leading to its latent phase. When reactivation occurs, movement of the virus in an anterograde fashion is seen towards superficial tissue that accounts for the characteristic symptoms of the infection.[4]

HZO accounts for 10-25% of all herpes zoster cases. This acute dermatomal infection is caused by reactivation of varicella virus in the ophthalmic division of the trigeminal nerve (V1).[5] An eruption of vesicular rashes in periorbital region and forehead occurs. Primarily, immunocompromised, older adults are targeted.[2-4] Complications develop in approximately 50% of patients with HZO, 5-31% of these patients report nerve palsies involving the ocular motor nerves. In contrast, ophthalmoplegia is very rare.[6,7] In various case reports HZO preceded ophthalmoplegia in 75% by a mean interval of 9.5 days and a range of 2 to 60 days, occurred simultaneously with ophthalmoplegia in 20%, and followed by 2 days the onset of ophthalmoplegia in only 5%. Our patient presented with HZO atleast two days after the onset of ophthalmoplegia. According to proposed theories, the ophthalmoplegia associated with HZO is because of direct cytopathic effect of the virus. It can also be due to an immunologic response to the virus which is reactionary in nature.[8,9] Ocular involvement is not uncommon in HZO. However, ocular pathology develops mainly in patients with involvement of the nasociliary nerve (Hutchinson's sign).[10] Eye manifestations also develop in approximately one-third of those without involvement of the nasociliary nerve.[11] Majority of the treated patients have a near complete reversal of ophthalmoplegia with a mean interval of 4 months as highlighted by various case series. Our patient also started showing signs of recovery with the start of extraocular movements demonstrated by moving his eye superiorly and lifting his ptotic upper lid within two weeks of treatment onset. Thus early recognition, treatment, and referral can prevent many poor outcomes.[12-14]

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