A misdiagnosed patient with an erythematous rash covering the half of his midface

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Ασθενής με ερυθηματώδες εξάνθημα στο ημιμόριο του μέσου προσώπου. Διαγνωστικοί προβληματισμοί

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DOI: 10.54936/haoms232133136

Case report
Αναφορά περιστατικού

SUMMARY: The aim of this article was to present the case of a 64-year-old male who had visited the emergency department of a tertiary hospital, and chiefly complained about a painful aggravated erythema on the right side of his face, bilateral periorbital swelling as well as redness and irritation in his ipsilateral eye. The patient described that the skin lesions had arisen 7 days before, and an out-of-hospital healthcare practitioner had initially attributed them to an acute and diffuse infection of his ipsilateral maxillary sinus. Pustules and crusts had covered the right half of patient's middle facial third, while conjunctivitis, keratitis, episcleritis, and ulcers on upper labial mucosa and hard palate were evident unilaterally too. Based on these clinical findings, what could be your diagnosis?

KEY WORDS: *herpes zoster, orofacial, maxillary division, trigeminal nerve, misdiagnosis*

ΠΕΡΙΛΗΨΗ: Σκοπός του παρόντος άρθρου ήταν να παρουσιάσει την περίπτωση ενός άνδρα 64 ετών που επισκέφθηκε το τμήμα επειγόντων περιστατικών τριτοβάθμιου νοσοκομείου και υπέδειξε ως αίτιο προσέλευσης επώδυνο και επιδεινούμενο ερύθημα στη δεξιά πλευρά του προσώπου του. Ο ασθενής προσδιόρισε ότι οι δερματικές αλλοιώσεις εμφανίστηκαν 7 ημέρες πριν, και ένας εξωνοσοκομειακός ιατρός τις απέδωσε αρχικά σε οξεία και διάχυτη λοίμωξη του ιγμορείου άντρου Κατά την επισκόπηση εμφάνιζε αμφοτερόπλευρο οίδημα στο πρόσωπό του, καθώς και έντονη ερυθρότητα στον δεξιό οφθαλμό του. Φλύκταινες, εφελκίδες, εσχάρες και κρούστες είχαν καλύψει το δεξιό μισό του μέσου τριτημορίου του προσώπου του, ενώ επιπεφυκίτιδα, κερατίτιδα, επισκληρίτιδα και έλκη στον σύστοιχο βλεννογόνο του δεξιού μισού του άνω χείλους και της σύστοιχης σκληρή υπερώας ήταν επίσης χαρακτηριστικές εκδηλώσεις. Με βάση αυτά τα κλινικά ευρήματα, ποια θα μπορούσε να είναι η διάγνωσή σας;

ΛΕΞΕΙΣ ΚΛΕΙΔΙΑ: *έρπης ζωστήρας, γναθοπροσωπική, διαίρεση της άνω γνάθου, τρίδυμο νεύρο, λανθασμένη διάγνωση*

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Paper received: 8/6/2022 - Accepted: 8/6/2022

Παρελήφθη: 8/6/2022 - Έγινε δεκτή: 8/6/2022

INTRODUCTION

A 64-year-old male visited the emergency department of Evaggelismos Hospital of Athens, and chiefly complained about a painful aggravated erythema on the right side of his face, bilateral periorbital swelling as well as redness and irritation in his ipsilateral eye (Figure 1). The patient described that skin lesions arose 7 days before, and an out-of-hospital otolaryngologist initially attributed them to an acute and diffuse infection of his ipsilateral maxillary sinus. Due to such a diagnosis, he had been treated with oral antibiotics and nasal decongestants.

The patient was examined by a maxillofacial surgeon who identified that the right half of patient's middle facial third had been covered by pustules and crusts. Ulcers on upper labial mucosa and hard palate were evident unilaterally too. A specialized ophthalmologic examination revealed conjunctivitis, keratitis, and episcleritis, without visual impairment (Figure 2). The skin manifestations initially consisted of both itchy macules and papules which subsequently formed vesicles. What is your diagnosis?

Answer. The clinical diagnosis of Herpes Zoster (HZ) was established by the maxillofacial surgeon (1st author), since the lesions were characteristically distributed along the branches of unilateral maxillary division of trigeminal nerve (Figure 3). The patient was also examined by a dermatologist, and his treatment comprised oral administration of valaciclovir 1 gr tid for 10 days, methylprednisolone 16 mg tid for 4 days and then taper for the next 4 days, ibuprofen 400 mg (tid for 5 days), and omeprazole per os 20 mg (bid for 5 days). Dexamethasone suspension eye-drops (tid. for 10 days) and antiviral acyclovir ointment (qid. for 10 days) were prescribed too. The lesions resolved without vision complications (Figure 4), but 13 weeks after the occurrence of viral infection, the patient mentioned neuropathic pain episodes on a daily basis, despite pregabalin intake.

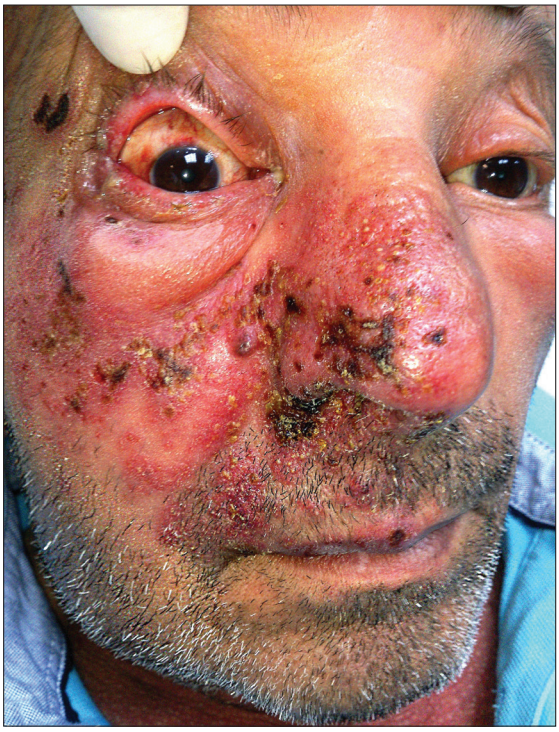

Fig. 1: Bilateral periorbital swelling and the right half of patient's middle facial third with erythema and covered by pustules and crusts.

DISCUSSION

The varicella-zoster virus (VZV) is a human alpha herpes virus, which is responsible two clinical entities; varicella (chicken pox) and herpes zoster (HZ) or shingles. In case of varicella skin infection, the virus is transferred along the sensory nerves to become latent in the sensory ganglia (8). The VZV remains in latent state, but when it is reactivated in the dorsal root ganglia, cutaneous and mucosal lesions of HZ appear along the dermatomes.

The incidence of HZ infection shows significant association with increasing age, since 99.5% of adults at age 40 years or older exhibit antibodies against VZV (5,13). Approximately 4% of individuals with HZ infection will experience relapse in their lifetime. The predisposing conditions involve malnutrition, (long-term) corticosteroid therapy, cytotoxic medications, radiotherapy, chemotherapeutic agents, diabetes, chronic obstructive pulmonary disease, malignant disease, and immune disorders (2). Reactivated maxillary HZ combined with neurotrophic keratitis has been reported after percutaneous block with alcohol, used for the treatment of trigeminal neuralgia, in the area of 2nd and 3rd branches of trigeminal nerve (1).

The clinical course of HZ is separated into three consecutive stages: the prodromal, acute, and chronic neuropathic (8). The prodromal/pre-eruptive stage presents as pain in conjunction with mild fever, headache, and dysesthesia. The acute stage features a rash in a unilateral dermatome; within 1 to 7 days the rash turns into pustule, which in turn dries and forms a painful crust after 14 to 21 days. The neuropathic stage follows with sharp, intense, radiating pain lasting about 1 to 3 months, but it may also persist for years and decades. Laboratory diagnosis of HZ may be established by cytological examination (Tzanck smear: Tzanck cells, intranuclear inclusion bodies, and numerous lymphocytes), fluorescent antibody staining technique (direct fluorescent antibody after testing of vesicular fluid or a corneal lesion), and PCR (2).

The trigeminal nerve is the most frequently affected of all the cranial nerves (18.5 to 22 %), followed by glossopharyngeal and hypoglossal nerves (6). In general, the V1 dermatome is the most frequently affected, while the V2 and V3 dermatomes (Figure 5) are usually less implicated (9). Even though rare, the complications of

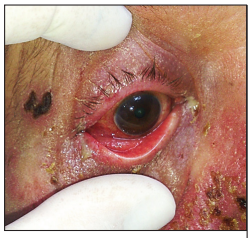

Fig. 2: Conjunctivitis, keratitis, and episcleritis, without visual impairment.

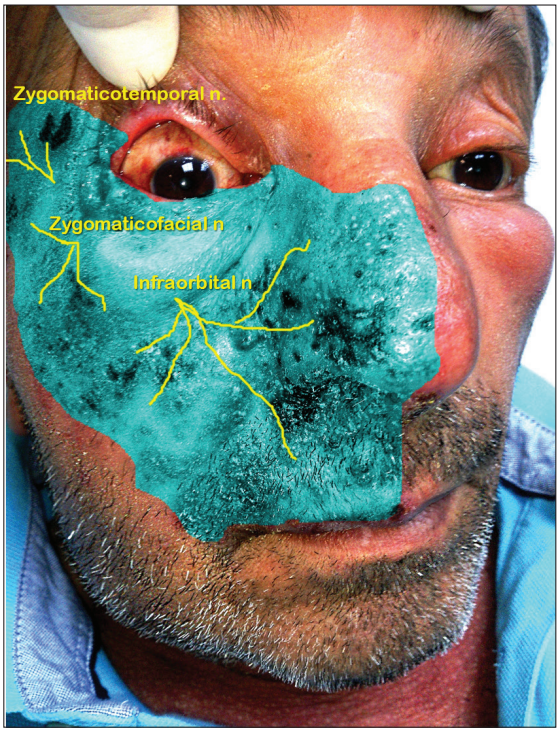

Fig. 3: Distribution of lesions along the branches of unilateral maxillary division of trigeminal nerve.

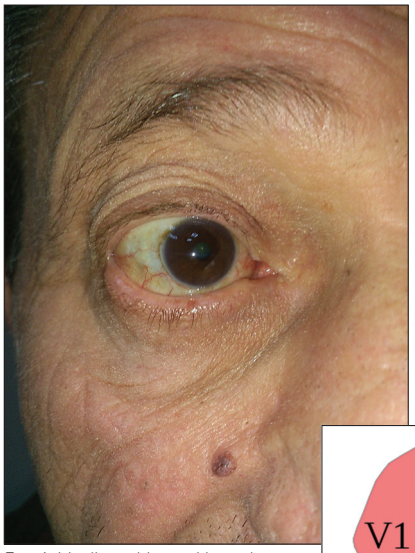

Fig. 4: Healing without skin and vision complications.

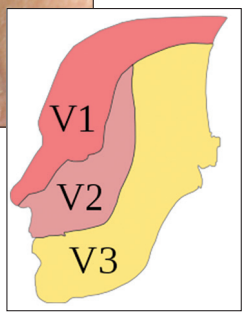

Fig. 5: DThe V1, V2, and V3 dermatomes.

maxillary HZ, arising in dentoalveolar complex, may include periapical lesions, calcified and devitalized pulps, root resorption of involved teeth, jaw osteonecrosis, and spontaneous tooth exfoliation (6). Local ischemia, pre-existing pulpal or periodontal inflammatory background, surgical trauma, and viral infection of specific cell populations may result in degenerative mechanisms on tissues (4). HZ of the maxillary division generates vesicles on the uvula and unilateral tonsil too. HZ oticus also named as Ramsay Hunt syndrome is a rare variant of facial HZ which involves otalgia, auricular vesicles, tinnitus, vertigo, loss of taste, and peripheral facial paralysis (11). In comparison to patients with Bell's palsy, those with Ramsay Hunt syndrome statistically demonstrated incresed risk to have severe and complete denervation with persistent synkinesis.

On the other hand, HZ ophthalmicus has been associated with the prodrome of tingling over the unilateral forehead region, whereas its specific manifestations may comprise severe ocular pain, corneal and profuse eyelid edema, conjunctival, episcleral, and circumcorneal conjunctival hyperemia, and photophobia (10). Keratitis, uveitis (chronic or recurrent), corneal scarring, glaucoma, cataract, episcleritis, corneal neovascularization, hypesthesia, and retinitis (high risk for severe visual impairment) are complications of HZ ophthalmicus that may occur too (10).

Hospital admission should be strongly considered for severe signs and symptoms, immunosuppressive status, disseminated herpes zoster affecting more than 2 dermatomes, extended facial bacterial superinfection, meningoencephalopathic manifestations, ocular involvement, and atypical signs (e.g. myelitis lesions) (8). Prompt diagnosis of HZ is vital, provided that timely initiation of antiviral therapy (acyclovir, famciclovir, orvalacyclovir) at early stages of the disease (within 72 hours of first manifestations occurrence) may contribute to the reduced severity of lesions and shorter healing time (12). Other medications being used in the total management include systemic steroids (hasten the resolution of acute neuritis), gabapentin, pregabalin, cyclic antidepressants, topical capsaicin or lidocaine ointment, and botulinum toxin injection (7).

In terms of prevention, the live-attenuated VZV vaccine (no longer available in the US) has contributed to a notable decrease in the incidence of primary varicella infection, and its efficacy may be maintained for 3 years after vaccination (3). The newer, recombinant vaccine seems to offer improved and longer-term protection than the older, single-dose, live-attenuated zoster vaccine (7). Either the recombinant or the live-attenuated vaccine are considered suitable for immunocompetent adults ≥ 50 years, but the recombinant one is more preferred (7).

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