Case Reports in Dermatology

Case Rep Dermatol 2024;16:102–107
DOI: 10.1159/000537901
Received: August 11, 2023
Accepted: February 15, 2024
Published online: April 10, 2024

Single Case

Thoracocervicofacial Purpura as a Presenting Symptom of Seizure Disorder: A Case Report

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Keywords
Thoracocervicofacial purpura · Valsava · Conjunctival hemorrhage · Nocturnal · Seizure

Abstract
Introduction: Postictal thoracocervicofacial purpura (PTP) is a rare clinical sign after a seizure episode and may be the only objective sign seen in patients who are unaware of their own seizure disorder. Moreover, it can be the only reason a patient seeks medical care after a seizure activity. ***Case Presentation:*** Herein, we report a 23-year-old, living alone and not known to have any medical illnesses, who presented to our emergency department complaining of asymptomatic purpuric facial rash extending to his neck and upper chest that started suddenly when he woke up that morning. Tongue and distal lateral thigh erosions were also noticed during physical examination, as well as conjunctival hemorrhage. In addition, the patient stated that he experienced similar prior episode of purpuric rash over the face, and disappeared uneventfully within 2 weeks. Blood work was remarkable for elevated urine creatinine (21,692 mg/L) and creatine kinase (1,207 mg/L). Given his clinical features and initial laboratory results, a diagnosis of PTP was made. ***Conclusion:*** Patients who present with petechial rash over the chest, neck, malar area along with conjunctival hemorrhage as an only complaint should be suspected to have or evaluated for seizure disorder.

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Introduction

There have been a variety of clinical signs and cutaneous manifestations observed following seizure, among them are automatism, hypersalivation, traumatic ecchymosis, cutaneous abrasions, choroidal venous congestion, subconjunctival hemorrhage, or tongue ulcerations. Thoracocervicofacial purpura is an uncommon postictal presentation and is mostly reported in the neurology literature but not to the same extent in the dermatology literature. It is hypothesized that it results from an increased intrathoracic pressure resulting in elevated venous pressure and extravasation of blood. Dermatologists and other healthcare professionals should be aware of this cutaneous phenomenon since lesions can be misdiagnosed as drug-induced eruption or viral exanthem [1]. Moreover, this clinical symptom could be the only manifestation of an underlying epileptic disease and should be in the differential for any patient presents with petechiae or purpura localized to the upper body [2].

Case Presentation

A 23-year-old male patient, medically free, presented to the emergency department complaining of a facial rash that started suddenly after he woke up over his forehead extending to his upper and lower eyelid, cheeks, nose, and neck. The rash was not itchy, painful, nor exacerbated by sunlight, but was associated with fatigue, drowsiness, tongue biting, falling of the bed, and painless redness of both his eyes. No history of head trauma, photosensitivity, lip smacking, involuntary hand movements, blinking. Three months prior, a similar episode of tongue biting, drowsiness, syncope, and purpuric rash happened, which healed spontaneously within 2 weeks. Family history is remarkable for psoriasis. As for his social history, he lives alone and was not witnessing any abnormal movement. His physical examination upon presentation showed stable vital signs. He had an abrasion over his tongue (shown in Fig. 1) and a small abrasion in the right distal thigh with well-circumscribed margins (shown in Fig. 2). Numerous non-palpable, non-blanching, non-tender petechiae were seen on the forehead, periorbital area, mid-face, maxilla, neck, upper chest, and shoulders during skin examination (shown in Fig. 3, 4). In addition, fundal examination was unremarkable, conjunctival examination revealed bilateral hemorrhages with a clear lens and cornea (shown in Fig. 4). His blood work showed unremarkable complete blood count with differential, coagulation, and liver profiles. Urine creatinine was 21,692 mg/L and creatine kinase of 1,207 mg/L. Referring to the previous clinical findings and blood work the diagnosis was suggested to be recurrent postictal thoracocervicofacial purpura by dermatology initially. The patient was reassured about the purpura and did not require any further dermatological treatment. Electroencephalogram and CT brain were inconclusive. The patient was referred to neurology for further assessment, given his clinical features and elevated CK levels, a diagnosis of epilepsy was confirmed and levetiracetam 500 mg was given to the patient to control his seizure along with continuing further investigations to detect the etiology of recurrent seizures.

Discussion

Postictal thoracocervicofacial purpura (mask phenomenon) is considered under-recognized uncommon secondary finding of epilepsy [1–8]. In relation to our presented case, postictal petechiae are characterized to be non-retiform, involving anterior chest, face, particularly periorbital areas, and neck, in addition to conjunctival hemorrhage [1, 2, 6, 9, 10]. Referring to the literature, no systematic reviews, cohort or cross-sectional studies were done as references for

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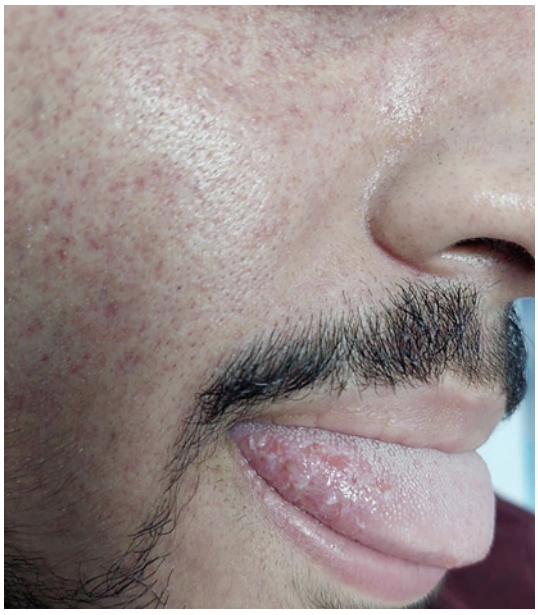

Fig. 1. Eroded tongue secondary to biting during ictal episode.

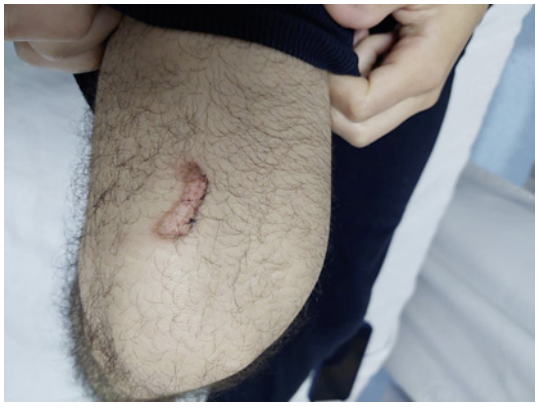

Fig. 2. Well-demarcated eroded plaque over the distal thigh.

this report, which supports the rareness of this phenomenon. Yet our argument, which was built according to similar case reports, divides the pathophysiology of purpura into two major categories; disruption in vascular integrity (such as trauma, infection, vasculitis, etc.), disorders of hemostasis (such as thrombocytopenia, platelet function abnormalities, coagulopathies, or disseminated intravascular coagulation). When applying the previous suggestions on our case, we discover that extravascular mechanism of unintentional prolonged increases intrathoracic or intra-abdominal pressure by at least 30–40 mm Hg (valsalva maneuver) caused by nocturnal convulsions [1, 2, 8–12] that results in extravasation of blood cells due to capillary damage, is the main event for causing epilepsy induced thoracocervicofacial rash [1, 2, 4–11]. Similarly, conjunctival hemorrhage follows the same pathophysiology of thoracocervicofacial purpura indicating the transmission of high intrathoracic pressure through choroidal vessels, causing them to burst, which perfectly aligns with the presentation of this case [5, 13]. In addition, the destructive effects of vasoactive mediators produced by neurons upon small capillaries are another proposed hypothesis for ictal purpuras [1, 2, 7, 9, 11]. De Souza et al. [7] reported similar clinical presentation to our case; however, purpuric lesions disappeared within 48 h. Our patient's petechial rash took 2 weeks to clear after the first ictal episode, and 3 weeks after the second episode as

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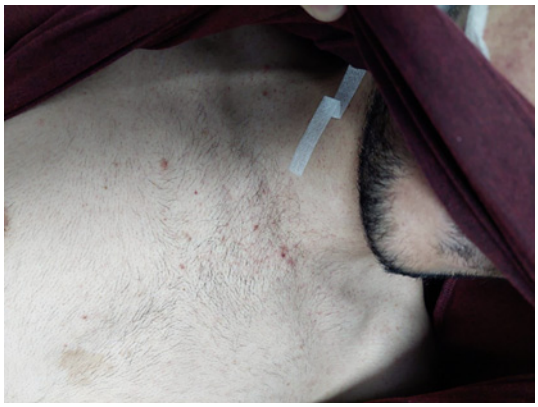

Fig. 3. Petechial rash over the upper chest.

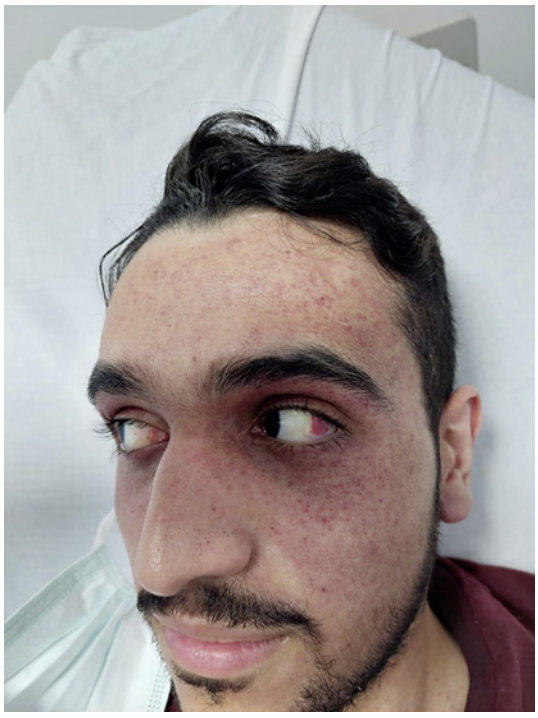

Fig. 4. Periorbital purpuric rash and conjunctival hemorrhage.

evident during outpatient dermatology follow-ups [7]. As mentioned earlier that postictal purpura tends to appear following nocturnal seizure rather than daytime seizure, Wali and Ziller [14] described a woman with periorbital purpura occurred after nocturnal ictal episode; however, it was witnessed by her husband. Our patient lives alone, hence, it was difficult to establish the diagnosis of epilepsy from the first ER visit. In both occasions, his skin rash was the main complaint. Confirmed diagnosis of epilepsy-induced thoracocervicofacial purpura is reached by exclusion of other purpuric pathologies by history and physical examination in addition to necessary laboratory workups. Our patient's tongue erosions secondary to bite during seizure and thigh erosions indicated traumas as external process during his ictal episode, while his purpuric rash and conjunctival hemorrhage indicate elevated intravascular pressure secondary to increase intrathoracic and intraocular pressure an internal process while seizing (shown in Fig. 2, 3, and 4). Neurology consultation and treatment of underling disease are crucial for controlling purpura recurrence in addition to supportive care since the disease is self-limited and complete

resolution of lesions will be achieved within days to weeks [2, 4, 6, 10, 11]. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000537901).

Conclusions

Making this benign rare phenomenon well recognized by medical practitioners and dermatologists is crucial for the rapid diagnosis of underlying epilepsy. As it could be the only presenting complaint a patient would consult a health care provider for after unwitnessed nocturnal convulsion. Referring to our previous arguments of postictal thoracocervicofacial purpura, we encourage other researchers to build a thesis of possible relation between first petechial presentation and nocturnal convulsions.

Statement of Ethics

The authors have no ethical conflicts to disclose. Written informed consent was obtained from the patient himself for publication of the details of their medical case and any accompanying images. The research was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. The study protocol was reviewed and approved by the institute's committee on human research named: the General Directorate of Health Affairs of Makkah Region. The date of approval: May 16, 2023. Information revealing the subject's identity is to be avoided.

Conflict of Interest Statement

The authors have no conflict of interest that is directly relevant to the content of this case.

Funding Sources

No sources of funding were used to assist in preparation of this manuscript.

Author Contributions

All authors made huge efforts in structuring this case report. Conception and design of the study: Waseem Alhawsawi. Data collection: Rama Halabi. Introduction and discussion: Basant Alzubaidy. Drafting the manuscript: Ammar Baksh. Revising the manuscript for critically for important intellectual content: Khalid Al Hawsawi. Patient care: Alhusain Alshareef. All authors approved the version of the manuscript to be published.

Data Availability Statement

All data that support the findings of this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

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