

Pearls & Oy-sters: Supraorbital neuralgia in lepromatous leprosy masquerading as SUNA

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Pearls

- Triad of continuous supraorbital and forehead pain in the sensory territory of supraorbital nerve, a tender supraorbital nerve, and complete but transient improvement in pain after supraorbital nerve block characterize supraorbital neuralgia.
- Supraorbital neuralgia can occur due to head trauma, tight-fitting helmet, and ill-fitting swimming goggles, but has never been described secondary to leprosy.
- One should look for hypopigmented skin patches at extensor aspect of elbow, gluteal region, and lower back and also palpate all superficial nerves in a patient with supraorbital neuralgia, especially in leprosy endemic regions.

Oy-sters

- Stroking a thick and tender supraorbital nerve in leprosy may trigger a burst of short-lasting periorbital pain mimicking short-lasting unilateral neuralgiform headache with cranial autonomic symptoms (SUNA).
- SUNA can be differentiated from the episodic bursts of pain that occur in supraorbital neuralgia by the lancinating character of pain, absence of Tinel sign, ability to trigger pain by touching periorbital skin, yawning, chewing, presence of lacrimation or redness with pain, and asymptomatic interpain interval.

Case report

A 24-year-old man presented with right forehead pain for 1 year. The pain frequently extended upwards to involve the right frontal and parietal scalp region. It was dull, continuous, and aching type with intermittent short-lasting spurts of bursting headache in the same territory. The patient noticed a swollen nerve in the right forehead region for 2 months. The swollen nerve was painful to touch. Stroking firmly on the nerve produced a brief but very severe pain in the eyebrow and right eye radiating up into the right scalp. This triggered pain was sometimes severe enough to produce lacrimation from the right eye. The pain usually lasted for a few seconds. There was no history of head trauma. He received a diagnosis of short-lasting unilateral neuralgiform headache with cranial autonomic symptoms (SUNA) and received lamotrigine, amitriptyline, gabapentin, and carbamazepine, but with no relief.

On examination, the patient had a visible, thick and firm right supraorbital and right greater auricular nerve. His ulnar, superficial radial, and common peroneal nerves were also thick and tender. The right greater auricular nerve showed a beaded appearance (figure). He also had multiple hypopigmented and hypoanesthetic skin patches in the extensor aspect of elbow, gluteal region, and lower back. Skin biopsy from one of the affected skin patches revealed noncaseating granuloma and acid-fast bacilli on Wade-Fite stain. The patient received multi-drug treatment for lepromatous leprosy and a brief period of treatment with oral prednisolone (1 mg/kg, tapered over 4 weeks). His skin patches disappeared in 3 months. The pain of

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(A) Thick, right supraorbital nerve. (B) Thick and beaded right greater auricular nerve. (C) Hypopigmented large skin patch on extensor aspect of left elbow.

supraorbital neuralgia also improved. The previous thickening of right supraorbital nerve was reduced.

Discussion

A triad of spontaneously occurring continuous nonthrobbing pain in the territory of supraorbital nerve, tender nerve in the supraorbital notch, and complete but transient relief from pain by supraorbital nerve block characterizes the supraorbital neuralgia.¹⁻³ Pain in supraorbital neuralgia is usually non-lancinating but it is accompanied by intermittent bursts of periorbital/frontal bursting pain that usually lasts for a few seconds.³ Supraorbital neuralgia is a rare headache. The most common cause of supraorbital neuralgia is forehead trauma and often, these patients will have stich marks above the eyebrow on the affected side. Other causes include tight-fitting helmets and ill-fitting swimming goggles (goggles migraine).^{4,5} However, it has never been described secondary to leprosy. Leprosy may have varied manifestations and therefore it is considered to be a great mimic in endemic areas.⁶

The triad for the diagnosis of supraorbital neuralgia requires supraorbital nerve block to demonstrate transient pain relief. We use nerve block only after failing with medical management. Our patient responded to the multidrug treatment for leprosy after failing a number of antiepileptic medications. Our patient had multiple thickened and tender nerves, suggestive of leprosy neuritis. Therefore, we also prescribed oral prednisolone for a 4-week period along with the antileprosy treatment, which might have enhanced the effect of antileprosy treatment. Leprosy is a disease of peripheral nerves caused by *Mycobacterium leprae*.⁷ It produces thickened and tender nerves. Inadvertent stroking of the enlarged nerve may produce severe lancinating pain in the territory of the nerve that is best explained by the Tinel sign. Therefore, it was not

the nerve trauma but the inflammation of supraorbital nerve secondary to leprosy that resulted in Tinel sign in our patient. Supraorbital neuralgia is usually not associated with lacrimation or eye redness. However, some patients report mild transient lacrimation on rare occasions.⁵ Our patient reported an event when he hit his forehead on his friend's helmet while riding pillion on a bike. He experienced very severe lancinating pain in the eye and forehead region with lacrimation. Possibly, this history of lacrimation from the affected side on a few occasions with episode of triggered pain and the history of intermittent spontaneous burst of periorbital/forehead pains of supraorbital neuralgia mistakenly led to the diagnosis of SUNA in our patient.

SUNA is characterized by multiple daily pain attacks in the eye/periorbital region that last for several seconds and are usually accompanied by lacrimation or redness of the affected eye. The pain attacks are spontaneous as well as triggered. The pain is in the form of single or multiple stabs of lancinating pains.⁸ There is no background continuous pain in SUNA, unlike the continuous pain seen in supraorbital neuralgia. The pain of SUNA is largely indistinguishable from the pain of trigeminal neuralgia of ophthalmic division. The supraorbital neuralgia, however, shares the distribution of pain with SUNA because supraorbital nerve is a terminal branch of frontal nerve, which is a branch of ophthalmic division of trigeminal nerve. The triggered pain in supraorbital neuralgia is always secondary to Tinel sign. In contrast, the pain in SUNA can be triggered by different activities like touching the eyelid or periorbital area, chewing, brushing teeth, yawning, or sometimes by talking.

Author contributions

V.K. Paliwal: conception and drafting of manuscript. S. Anand: analysis of clinical data, drafting of manuscript. A.S. Rai: acquisition of clinical data and revised the

manuscript. R. Chiroliya: wrote figure legends and revised the manuscript.

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Disclosure

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