



Atypical Trigeminal Trophic Syndrome: A New Cause of Facial Ulceration?

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Clinical Image

We present a case of a 56-year-old Caucasian male patient, who presented to our specialist psychodermatology clinic, complaining about severe chronic facial pain, bilateral ulceration and scarring, following the mandibular nerve distribution. All this condition started 6 years ago. The affected areas included left and right cheeks, left post-auricular area and chin. The triggering factor appeared to be an episode of shingles. Before his referral at our clinic, his skin lesions were attributed either to dermatitis artefacta or acne excoriée. He was previously treated with antipsychotic medication and psychotherapy, with no significant improvement in his condition.

Differential diagnosis of facial ulcers included malignancy, infection, vasculitis, pyoderma gangrenosum and psychodermatological disorders (dermatitis artefacta, trigeminal trophic syndrome, atypical trigeminal trophic syndrome and acne excoriée).

Swabs from active ulcers were taken and were negative for pathogens, excluding infection. Blood results revealed microcytic anaemia, with no underlying cause. Biochemical check for pruritus, including liver function, urea and electrolytes, thyroid function, syphilis serology, was negative. Vasculitis serology was negative. A skin biopsy was performed showing only scarring of the epidermis, excluding malignancy and pyoderma gangrenosum. Magnetic resonance cranio-facial imaging was also performed showing mild chronic inflammation of the facial sinuses. Finally, neurophysiology, including electromyogram, revealed bilateral trigeminal nerve dysfunction on both trigeminal nerves, which was in favour of atypical trigeminal trophic syndrome.

After previous treatment with antipsychotics and psychotherapy with limited efficacy, the patient had a lengthy consultation with pain services in association with our multidisciplinary team and he was started on aripiprazole 5 mg daily, morphine sulphate (MST) 40mg twice daily, pregabalin 300 mg twice daily, duloxetine 60 mg twice daily, oxcarbazepine 150 mg twice daily and topical lidocaine patches.

Trigeminal Trophic Syndrome (TTS) is a rare but important cause of facial ulceration characterised by ulceration, anaesthesia and paraesthesia following the distribution of trigeminal syndrome [1,2]. Injury to the trigeminal nerve, central or peripheral, is the main causative agent. The resulting intractable dysaesthesia leads to self-mutilating behaviour resulting in ulcers [1-3]. Diagnosis is made clinically and neurophysiological studies can be helpful in evaluating function of the trigeminal nerve [4] TTS is a unilateral condition, typically responding well to antidepressants or antipsychotics [1,2].

Nevertheless, our patient presented with an atypical, bilateral condition that was highly refractory to established treatments. We believe this patient has a clinically new entity, named

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Figure: (A) Patient with ATTS before treatment. (B) Patient with ATTS after-treatment.

as atypical trigeminal trophic syndrome (ATTS) [5], which we are seeing in an increasing number of patients in our tertiary psychodermatology clinic. In ATTS, present with bilateral disease and this is confirmed by nerve conduction studies. Skin involvement is extensive and may fluctuate in severity. ATTS is highly refractory to treatment and an obsessive-compulsive element often develops, which is much less a feature of TTS. We also believe that patients with ATTS are particularly difficult to treat even with agents that are typically efficacious in TTS. Clinicians should be aware of this condition in order to ensure that patients are appropriately referred to an experienced, multidisciplinary team to manage this mutilating disorder (Figure 1A and B).

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