

Complex Regional Pain Syndrome of the Upper Limbs Caused by Facial Pain

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Polyacrylamide hydrogel is a widely used filler material in cosmetic procedures performed on the face and breasts. Recently, however, complications including inflammation, deformity, and pain have been reported. The present article addresses unregulated materials/products injected as dermal fillers. The authors report a case involving a 29-year-old woman who developed severe facial pain after undergoing a cosmetic procedure with injectable triamcinolone and hyaluronidase. Two months later, the pain spread to her upper and lower limbs, and abdomen, which eventually led to the development and diagnosis of complex regional pain syndrome (CRPS) in the upper limbs. The authors hypothesize that CRPS in the upper limbs was responsible for the facial pain through sensitization of third-order neurons and the trigeminal nucleus caudalis extending to the upper cervical segments.

Keywords: Complex regional pain syndrome, Dermal filler, Facial neuralgia, Facial pain, Hyaluronic acid

Introduction

Polyacrylamide hydrogel, consisting of polyacrylamide and 97.5% water, is commonly used in cosmetic procedures as a permanent filler material [1]. Although Polyacrylamide hydrogel has a low complication profile, adverse events such as infection, inflammation, lumpiness, gel indurations, and pain, have been reported [2]. It is believed that there may be an increase in incidence of these complications when unregulated materials/products are used in cosmetic procedures.

The present article describes a case involving a 29 year-old woman, who experienced severe facial pain after undergoing a facial cosmetic procedure. The pain involved radiation to the limbs, and abdomen, which eventually resulted in the development and diagnosis of complex regional pain syndrome (CRPS).

Case Report

A 29-year-old female presented with pain in her nose, forehead, abdomen, and both hands, and feet. Two years previously, she received an injection, administered by and aesthetic clinician, of materials/products in her nose that were not approved by the Korea Food & Drug Administration. One-year post injection, she underwent a minor deformity correction procedure performed by the same physician, in which 50 mg of injectable triamcinolone was used. Three months later, she was reinjected with 1500 IU of hyaluronidase. When triamcinolone was injected, she reported persistent mild pain after the procedure. She scored the pain as 2 or 3 on a visual analogue scale (VAS; 0 = no pain, 10 = severe pain). When 1,500 IU of hyaluronidase was injected, the patient reported severe pain immediately after the injection, which was scored as 8 or 9 on the VAS; she characterized the pain as a sharp and tearing, which spread from her nose to her forehead. Subsequently, the pain occurred intermittently throughout the day, accompanied by flushing of the forehead. The patient could not brush the hair on the front area of her scalp due to allodynia.

Two months later, the pain spread to her four extremities, and to her abdomen. The pain in her extremities was characterized as prickly and electrical sensations especially in her hands and

feet, whereas in her lower abdomen, it was characterized as tearing and stabbing sensations, scoring a 7 or 8 on the VAS. The pain was evoked even by gentle touch. At the same time, pale to dark-red discolorations appeared on both her of hands. Her medical history was non-contributory, and non-steroidal anti-inflammatory drugs were prescribed to control the pain. Otherwise, she always wore gloves and thick socks, because of cold sensation in both her hands and feet.

On physical examination, both of her hands felt cold and wet bilaterally. Her wrists were edematous bilaterally. The patient's symptoms are summarized in Table 1.

Laboratory investigations, magnetic resonance imaging (MRI) of the head and face, thermography on the face and body, and three-phase bone scintigraphy on the hands and feet were conducted upon visiting the author's clinic. The results of the laboratory tests were negative for anti-nuclear, anti-RNP, anti-centromere, and anti-Scl-70 antibodies. All other test results, including levels of anti-double-stranded DNA, complements 3 and 4, and rheumatoid factors, were all within normal ranges. The erythrocyte sedimentation rate and C-reactive protein level were also found to be within normal limits. MRI of the head and face was performed to determine alternative causes of pain, but revealed no inflammation or abnormal significant findings. In thermography, the mean ($\pm$ SD) temperature of the patient's fingers and toes were $23.19 \pm 0.25^{\circ}\text{C}$ and $22.73 \pm 0.02^{\circ}\text{C}$, respectively. The temperature of the volar side temperature of the hand and foot of the patient was $25.14 \pm 0.65^{\circ}\text{C}$ and $25.18 \pm 0.12^{\circ}\text{C}$, respectively (Fig. 1A&B) [3]. Relative hypothermia in both limbs was revealed bilaterally. Consequently, it was impossible to compare with a normal, non-involved side. Three-phase bone scintigraphy revealed an increase in blood flow in the delayed phase in both wrists and the small

Table 1. Summarized pain-related symptoms of the patient who presented to the our clinic

Location	Symptoms (s)	Sings
Face	Nose and forehead: sharp and tearing pain Forehead: flushing	Allodynia with brushing hair at frontal area
Upper limb	Hand: pricking, electrical like pain, cold sensation, color change from pale to dark-red Wrist: edema	Hand: hyperesthesia evoked by touch Thermography of hand: temperature of volar side of hands, fingers revealed a lower (>3°C) compared with 63 healthy adults Hand: cold and wet Wrist: delayed uptake on the three-phase bone scan, edema
Lower limb	Foot: pricking, electrical like pain, cold sensation,	Thermography of feet: volar side of feet and toes revealed a lower (>3°C) mean temperature compared with 63 healthy adults
Abdomen	Stabbing, tearing pain	

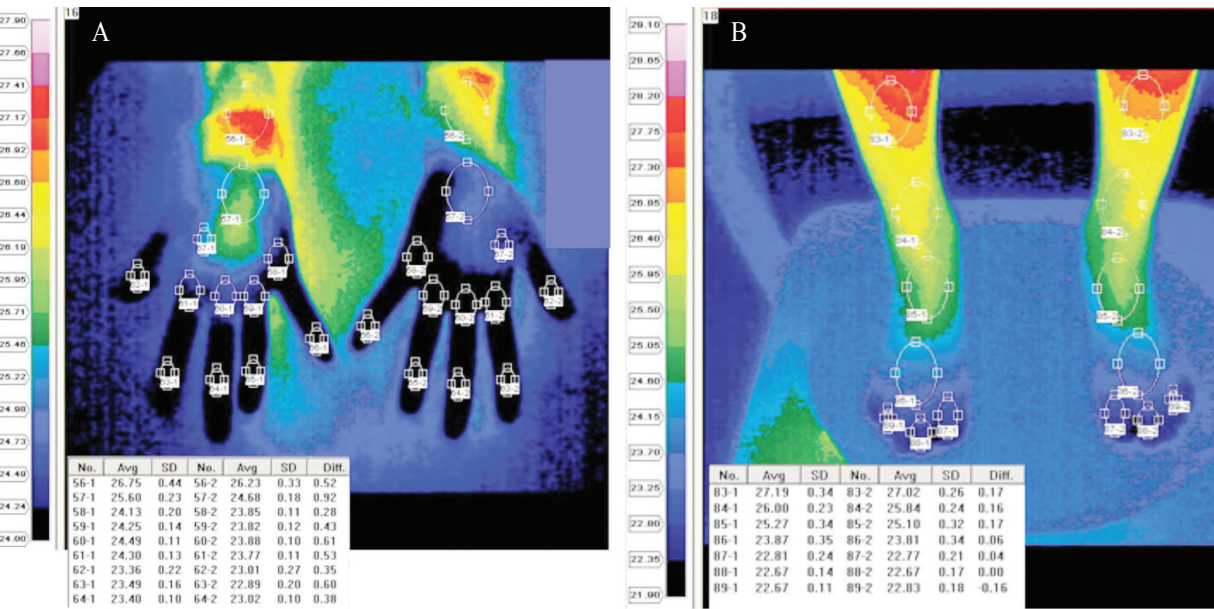


Fig. 1. Thermography of the hands and feet. (A) The mean ($\pm$ SD) temperatures of the fingers and the volar side of the hands were $23.19 \pm 0.25^{\circ}\text{C}$ and $25.14 \pm 0.65^{\circ}\text{C}$, respectively. (B) The mean ($\pm$ SD) temperatures of the toes and the volar side of the feet were 22.73 ± 0.02 and $25.18 \pm 0.12^{\circ}\text{C}$, respectively.

joints of both hands (Fig. 2A&B).

In reference to the 2004 budapest International Association for the study of pain (IASP) CRPS

diagnostic criteria, pain, color changes, wet hands, edematous wrist in upper limbs, fulfill at least one symptom of three categories. In previous study,

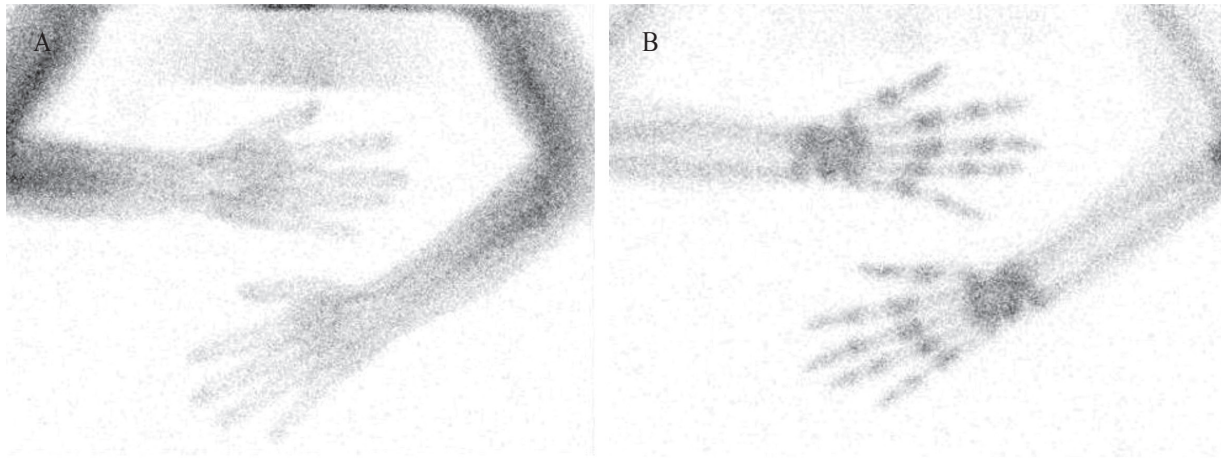


Fig. 2. Three-phase bone scan revealed uptake in both wrists in the delayed phase. (A) blood pool phase; (B) delayed phase.

hyperesthesia, allodynia of the arm, relative hypothermia of the fingers and volar side of hand exhibiting a lower mean temperature ($>3^{\circ}\text{C}$) were compared in 63 healthy adults [3]. Blood flow increases in the hand in delayed phase fulfilled at least one sign of two categories. In the present case, a diagnosis of CRPS in the upper limbs was ultimately established, and the patient was admitted to hospital for four weeks. During admission, continuous cervical epidural block (CCEB) for 7 days was performed three times. Ultrasound (US)-guided stellate ganglion block (SGB) with 0.5% lidocaine (5 mL) was also administered four times during the final week of admission, alternately between the left and right sides. Caudal blocks were performed four times (once per week) for lower limb pain during admission. The pain intensity decreased to a VAS score of 3 or 4 and the cold sensation in the patient's hands improved.

At discharge, the patient was prescribed pregabalin twice per day (300 mg/day), and tramadol twice per day (150 mg/day), combined with acetaminophen (1,300 mg/day). During follow-up, the patient reported tingling and

pricking sensations on her nose and in her extremities, with a pain score of 3 or 4 on the VAS; however, the pain in her abdomen had subsided.

Discussion

In the present case, we found that repeated cosmetic procedures performed on a female patient for the correction of a mild nose deformity led to the development and diagnosis of CRPS in the upper limbs owing to facial pain that spread to the upper limbs, and neuropathic pain in her abdomen and lower limbs. CCEB combined with US-guided SGB and caudal blocks, along with pregabalin twice per day (300 mg/day), and tramadol twice per day (150 mg/day), combined with acetaminophen (1,300 mg/day) reduced her pain to a score of 3 or 4 on the VAS.

Systemic corticosteroids [4] or local corticosteroids [5] have been reported to be effective in treating orofacial granulomatosis caused by injections of cosmetic materials such as silicon, hyaluronic acid, collagen, and methacrylate. Hyaluronidase is used for the

treatment of allergic reactions or to correct injection of hyaluronic acid in the wrong anatomical location(s) [6]. In the present case, injections of triamcinolone and hyaluronidase were administered to correct a deformity on the patient's nose, which caused severe facial pain. The exact mechanism of severe pain could not be fully explained; however, nerve injury or tissue damage during injection may have been responsible, leading to neuroinflammation. First, if nerve damage occurred, spontaneous neural activity and ectopic discharge to mechanical stimuli develop at the site of nerve injury, and central sensitization is dependent on signal transmission from these nociceptors [7]. Second, release of inflammatory mediators from non-neuronal tissue or inflammatory cells caused by tissue injury provoke pain and hyperalgesia, and cause peripheral nociceptor sensitization [8,9].

A diagnosis of CRPS is dependent on signs and symptoms. Diagnostic criteria were presented at the 2004 IASP conference (held in Budapest, Hungary) that demonstrates robust statistical sensitivity and specificity. Supportive diagnostic tests include three phase bone scan and thermography. Three phase bone scan demonstrated a sensitivity of 44%, a specificity of 92%, and a positive predictive value of 61% [10]. In the early stages of CRPS, blood flow increases in the perfusion or pooling phases of the affected limb(s), and bone resorption of radioisotope increases in the delayed phase. In the long-term, after disease onset, there is no increase although a possible decrease- in blood flow in the perfusion or pooling phases of the affected limb (s) [11]. Thermography is used to compare differences in body temperature between the affected limbs and normal parts. A temperature difference $>1^{\circ}\text{C}$ has recently been accepted for the diagnosis of CRPS. However, skin temperature differences between

bilateral limbs $\leq 1^{\circ}\text{C}$ have been reported in 44.3 % of patients. Therefore, although thermography is useful for diagnosis, it not a sufficient test [12]. In the present case, thermography revealed similar temperatures on bilateral limbs, because the upper and lower arms were involved symmetrically. This particular finding is still significant when compared with a previous report describing thermographic patterns in the hands and feet of 63 healthy adults. The mean temperature of the volar side of the hands, fingers, volar side of feet and toes were lower by at least 3°C than mean temperature [3]. These were one of the criteria for the diagnosis of CRPS.

The pain experienced by this patient, from the face to the limbs and abdomen, appear to be similar to that in a 22-year-old migraine patient who experienced from stabbing pain in her limbs and abdomen, followed by burning pain lasting 48 h [13]. A possible hypothesis explaining the spread of pain to other parts of the body in a migraine patient is sensitization of a third-order neuron, which transfers convergent information from the trigeminal ganglion and upper spinal dorsal horn [14,15]. The trigeminal nucleus caudalis extends to the upper cervical segments, whereas the lateral cervical nucleus has many afferent synapses that extend into other parts of the brain system. The lateral cervical nucleus also receives input from other body parts, such as the trunk and limbs [16]. These phenomena may explain the spread of CRPS from the trigeminal area to the upper limbs and neuropathic pain to the lower limbs and trunk of the patient.

An SGB is effective in treating CRPS in the head and neck. In addition, SGB is known to reduce pain in the face and arms [17]. A high thoracic epidural block was demonstrated to be as effective as SGB in alleviating pain in the head, which is innervated by the trigeminal nerve [18].

Conclusion

Unregulated material used as dermal filler on the face can cause nose deformities. Repeated traumatic procedures for the correction of this deformity may lead to CRPS in the face and spread to the upper limbs. Interventions, such as CCEB and cervical sympathetic block were effective in reducing the intensity of this pain.

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