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Painful Submandibular Hypertrophic Scar as a Presentation of Post-Traumatic Neuropathy: Case Report

Cicatriz Hipertrófica Dolorosa Submandibular como Presentación de una Neuropatía Post-Traumática: Reporte de Caso

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ABSTRACT: Painful hypertrophic scars (PHS) are benign fibrous overgrowths of scar tissue that manifest a subjective symptom with a pathophysiology poorly understood and an absence of a gold standard treatment, representing a challenging condition that can dramatically affect the patient's quality of life. A 16-year-old patient present pain intensity of 10 on the visual analog scale secondary to a PHS in the submandibular region. Treatment was carried out through three sessions of peri and sublesional local infiltrative anesthetic combined with posterior corticosteroid infiltration. After 7 months the patient was discharged, resuming all daily activities and without recurrence of pain during one year of follow-up. PHS have characteristics of neuropathic pain and may resemble a post-traumatic trigeminal neuropathy. Considering the unique complexity of each case and the multiple options available for its management, this case exposes an effective, simple, and non-surgical treatment showing successful results.

KEY WORDS: facial pain, hypertrophic scar, painful hypertrophic scar, mandibular nerve injuries, cicatrix; case reports.

INTRODUCTION

Scarring is a natural part of dermal healing processes involving inflammation, proliferation, and remodeling (Jalali & Bayat, 2007). Scars consist of networks of fibrous collagen tissue laid down in response to dermis injury (Jalali & Bayat, 2007). The inflammatory stage of wound healing is prevalent over the first 48 to 72 hours, overlapped by the proliferative stage, which is seen during the first six weeks, and the maturation stage can last throughout 1 to 2 years (Saddawi-Konefka & Watson, 2019).

Hypertrophic scars (HS) and keloids are benign fibrous overgrowths of scar tissue that result from abnormal wound healing following trauma, burn, surgery, or severe acne. These lesions can occur at any age and are characterized by a red, firm consistency to palpation and raised appearance (Wang *et al*., 2017; Cai *et al*., 2020; Leszczynski *et al*., 2022). In these cases, abnormal fibroblasts fail to undergo apoptosis and continue to produce connective tissue beyond the period expected for normal scars. Overexpression and inadequate regulation of growth factors, such as transforming growth factor beta (TGF-β), vascular endothelial growth factor, and connective tissue growth factor, may also play a role in altered tissue formation; there is a loss of balance between the production and subsequent degradation of collagen, where there is a higher production of collagen and lower levels of degradation (Viera *et al*., 2012).

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Establishing a clear differentiation between these two forms of benign fibrous overgrowths is challenging. The clinical diagnosis may not agree with the pathological diagnosis, with many cases having been registered where the scar bears the growth and histologic features of both HS and keloids (Ogawa *et al*., 2019). Furthermore, there are tools to objectively diagnose HS and keloids, such as the one presented by the Japan Scar Workshop (JSW), the JSW Scar Scale (JSS), developed in 2015 which involves scoring the risk factors and presents symptoms, allowing a simple use even for physicians who are not accustomed to these pathological scars (Ogawa *et al*., 2019).

Due to local stretching in the head and neck region, HS is conspicuous and difficult for patients to conceal (Wolfram *et al*., 2009; Ogawa *et al*., 2019). The location, size, and depth of the lesion, the patient's age, systemic factors, and previous response to treatment can determine the best strategy to manage this condition (Viera *et al*., 2012; Ogawa *et al*., 2019).

Subjective symptoms, such as a painful hypertrophic scar (PHS), can significantly affect a patient's quality of life. The prevalence of chronic post-operative pain is around 30-50 % after surgical procedures involving nerve transection or involving a high probability of nerve injury. The pathophysiology of PHS is poorly understood, and pain can be attributed to the stimulation of nerve endings by the local environment, resulting from hyperemia and scar growth during hyperplasia. The problem of painful scars can involve both intraneural and extraneural structures, thus requiring a systematic approach to the diagnosis and treatment of this neuropathic pain condition. The strained scar tissue localization, tissue stiffness, and its rebound effect on nerve tissue might be an indirect cause of neuropathic pain, although the specific mechanisms remain unclear (Xiao *et al*., 2018; Eker *et al*., 2019; Abd-Elsayed *et al*., 2022).

Examination of the painful scar includes visual inspection, palpation, mobilization, and diagnostic injection of local anesthetic in areas of allodynia or hyperalgesia, as appropriate. Currently, the literature describes as alternatives for the treatment of PHS the use of topical agents, like anesthesia and/or corticosteroids; pressure therapy; local drug injection, with corticosteroid being the most widely used; cryotherapy; laser therapy; surgical treatment; and post-surgical radiotherapy (Jalali & Bayat, 2007; Wolfram *et al*., 2009; Viera *et al*., 2012; Zhang *et al*., 2018; Saddawi-Konefka & Watson, 2019; Ogawa *et al*., 2019; Cai *et al*., 2020; Bao *et al*., 2020; Sun *et al*., 2021; Abd-Elsayed *et al*., 2022). In recent years, combination therapy has also been closely monitored, although some studies have found that combination therapy has shown good efficacy and safety (Cai *et al*., 2020; Bao *et al*., 2020). However, the most effective and safest combination for treating PHS has not yet been determined (Cai *et al*., 2020; Bao *et al*., 2020).

Therefore, this case report aims to describe a PHS after submaxilitis surgery and its respective treatment.

MATERIAL AND METHOD

A 16-year-old patient was referred by the maxillofacial surgery service with the chief complaint of pain in the submandibular region following two years of evolution. The pain began after left submaxilitis surgery, resulting in an extensive reddish and rigid scar as a sequel (Fig. 1). This scar scored 12 points on the JSW Scar Scale, being classified as an HS. Pain intensity was 10 on the visual analog scale (VAS), with features of dysesthesia, a burning painful baseline, and paroxysmal pain outbreaks that occurred when moving the jaw or exercising. Also, the pain caused changes in the patient's diet, as well as a significant negative social impact. Upon physical examination, pain is noted on palpation of the HS. Administration of infiltrative local anesthesia (mepivacaine 3 %) in the peri and sub-cicatricial zone resulted in a total reduction of pain, which finally confirmed the diagnosis of PHS. Furthermore, written informed consent was provided by the patient and her tutor to publish the case details and associated images for scientific and academic purposes.

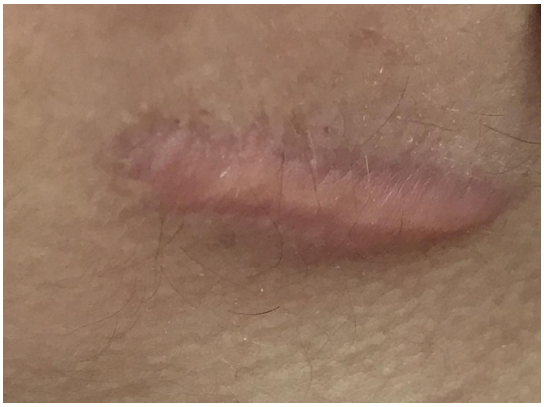

Fig. 1. Painful, reddish, and rigid hypertrophic scar secondary to left submaxillitis surgery. At the time of diagnosis, pain scored 10 points on the visual analog scale, associated with dysesthesia, basal burning, and outbreaks of paroxysmal pain.

RESULTS

In the present case, the initial therapeutic decision was based on the following characteristics: The high intensity of pain; the location of the lesion; pain triggered due to mandibular movement and touch in the area; functional deterioration of the jaw and difficulty feeding; and significant decrease of the patient´s quality of life, which merited rapid and effective management. Based on the above, treatment was started with 3% mepivacaine as a local infiltrative anesthetic (25G dental needle) in the peripheral and deep PHS area. Then, 1 mL of triamcinolone hexacetonide was infiltrated as a corticosteroid in the same location (infiltrated with a 25G hypodermic needle). Acetaminophen 1g every 12 hours was indicated as postoperative management for the first five days. After two weeks of follow-up, the patient reported a 50% reduction in pain intensity (VAS 5). The infiltrative procedure was repeated and monitored again after three months. At follow-up controls, the patient presented a VAS 3 with changes in the visual appearance of the HS (Fig. 2) and less adherence to deep layers.

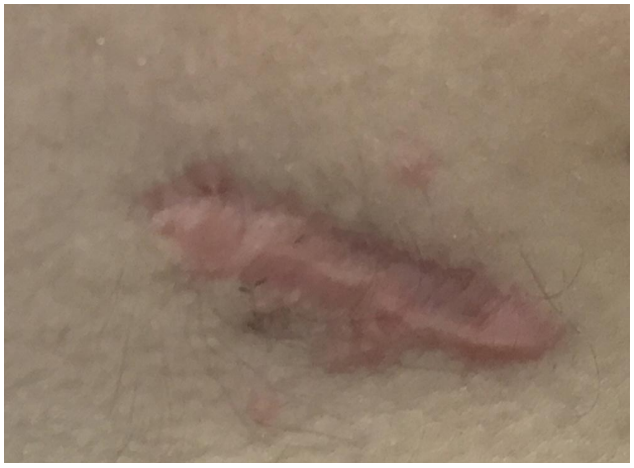

Fig. 2. Painful hypertrophic scar after three months of treatment, decreasing pain intensity to 3 points in visual analog scale. Visually, it is more deflated, less attached to deep layers, and less reddish.

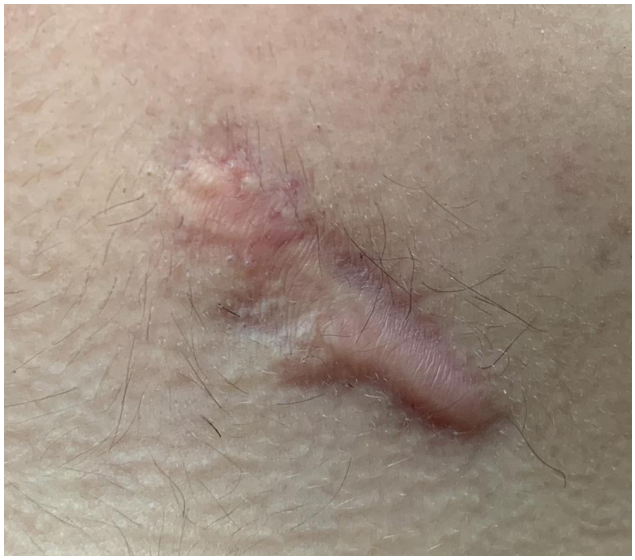

Fig. 3. Painful hypertrophic scar at four months of treatment, without spontaneous pain and painful response to manipulation or stimulation. Visually flat throughout its length and pale in color.

Furthermore, the patient presented less difficulty performing mandibular movements, resuming physical exercise, and engaging in regular social activities. A third infiltration was performed following the same protocol during this last session. Subsequently, at the time of the monthly control, the patient reported a VAS 0, the scar was observed pale in color (Fig. 3), and there was no painful response to manipulation or stimulation. No treatment or procedures were performed during this session. Finally, after the seventh month of starting treatment, the patient resumed all daily activities with no recurrence of pain and, thereby, was discharged. To monitor the case, the patient was contacted by telephone one year after discharge, maintaining good results of the treatment.

DISCUSSION

HS and keloids are benign fibrous growths of scar tissue that can cause pain and dysfunction, affecting patients' quality of life. The clinical differences between these two entities are not always simple to perceive, so the differential diagnosis could be complex. Table I compares the main characteristics of each one.

Although the pathophysiology of abnormal scar pain has not been fully described, it is attributed to the stimulation of free nerve endings (Xiao *et al*., 2018; Eker *et al*., 2019). These nerve endings, mainly from C fibers, deliver a neuropathic pain component to the clinical presentation. Thus, many of the symptoms are described as dysesthesia, allodynia, burning or paroxistic pain, or other neuropathic signs. These characteristics closely resemble those found in post-traumatic trigeminal neuropathies (PTTN) of the orofacial region. Many of the causes of HPS are traumatic or secondary to surgeries, as in the present case, like in PTTN. In the same sense, when reviewing the literature regarding treatments performed HPS cases, some are consistent with the treatments proposed for PTTN, which suggests a painful

Table I. Clinical features of hypertrophic scars and keloids.

Hypertrophic Scar	Keloid
• Do not grow outside the area of the original wound.	• Extend beyond the borders of the original wound.
• Have a low recurrence rate.	• Tend to recur after excision.
• Poorly associated with darker skin types.	• Associated more with darker skin types.
• Predominance of fine collagen fibers and heavy myofibroblast infiltration, secondary to aberrations in both extracellular matrix metabolism and hemostasis.	• Contain thick and uniformly stained collagen fibers that are called keloidal or hyalinized collagen and low numbers of myofibroblasts.
• Characterized by dermal nodules that are composed of increased numbers of collagen bundles that run in different directions.	• Collagen bundles are virtually nonexistent, and the collagen type I and III fibers lie in haphazardly connected loose sheets randomly oriented to the epithelial surface.
• No genetic association.	• Linked with an autosomal dominant pattern of inheritance.

pathophysiology shared between these entities. In this case, the submandibular region is sensitively innervated by the C2 and C3 nerves. Therefore, it could be a post-traumatic neuropathy of these nerves. However, the surgery that generated the PHS was performed in the submandibular gland, innervated by the lingual nerve, the terminal branch of the trigeminal nerve. Therefore, in this specific case, we could be facing a combined post-traumatic neuropathy.

As reviewed, corticosteroid infiltration has been a widely used minimally invasive therapy. The mechanism of action is the inhibition of fibroblast growth and decreased levels of alpha-2 macroglobulin, leading to collagen degradation. When used as a monotherapy, corticosteroids have a recurrence rate of 9 % to 50 % (Jalali & Bayat, 2007; Bao *et al*., 2020; Sun *et al*., 2021; Abd-Elsayed *et al*., 2022). For this reason, whenever possible, long-term follow-up is considered relevant, to verify the positive result of the therapy or evaluate the need for a new intervention. In this sense, it has been recommended to add, prior to infiltration of corticosteroids, topical anesthetic, such as gel (tetracaine) or cream (lidocaine or prilocaine), or infiltrative local anesthesia (xylocaine at 1 % or 3 % mepivacaine) (Wolfram *et al*., 2009; Wang *et al*., 2017; Ogawa *et al*., 2019), to avoid neuronal sensitization further and of course pain due to the procedure. The use of a fine needle for the procedure (30G, 27G, or 25G) is also recommended, avoiding a further increase of trauma in the area (Wolfram *et al*., 2009; Wang *et al*., 2017; Ogawa *et al*., 2019), and these could be combined with other conservative treatments, such as the administration of oral medication (ex: post-operative analgesics) (Wolfram *et al*., 2009; Wang *et al*., 2017; Ogawa *et al*., 2019). As clinical recommendations, caution and precision are also mentioned in the literature when infiltrating the corticosteroid, ensuring the drug enters the dermis. If the depth is too shallow, the treatment effect is poor; injected into the subcutaneous tissue may cause adverse reactions such as atrophy of the subcutis and telangiectasia (Cai *et al*., 2020). The rising pressure caused by injecting the hard mass may also cause pain. Instead, the needle should enter the scar from its margin with the normal skin and target either the deepest part of the scar, which is softer than its central core, or the most heavily inflamed part of the scar at the junction between the normal skin and the scar. Additionally, any surrounding active acne lesions should be avoided in the facial and cervical regions. If pathological scars are among the active acne lesions, these should be the priority of treatment (Ogawa *et al*., 2019).

Botulinum Toxin A (BTA) injections were reported as an alternative to scar pain reduction method. BTA is hypothesized to directly exert analgesic properties by reducing the release of mediators like substance P, glutamate, and calcitonin gene-related peptide (CGRP) in addition to immobilize the local muscles and reduce the skin tension caused by muscle pull (Uyesugi *et al*., 2010; Shaarawy *et al*., 2015; Schuler *et al*., 2019). Although, it is a more expensive treatment than corticosteroid injections. Another therapy described in the literature to manage HPS is the peri- and sub-scar infiltration of 100 mg of diclofenac sodium and 50 mg of 0.5% lidocaine in 10 mL of saline, presenting a good response at 8 - 12 weeks (Eker *et al*., 2019). In reviewing the scientific literature, most studies on PHS are case reports that agree on the diagnostic difficulty and describe different therapeutic alternatives. Table II summarizes some of the therapeutic options mentioned in the literature. It should be noted that among these therapies, the pharmacological treatment described for neuropathic pain or PTTN needs to be better defined or considered.

Table II. Therapeutic tools described for the management of hypertrophic and keloid scars.

Topical agents	Widely used, with non-steroidal anti-inflammatory drugs and corticosteroids being the most widely used. These can be applied as ointments, creams, gels, and skin patches. Sometimes they are associated with occlusive therapyperformed on or around the scar, or used with silicone gel sheeting.
Local drug injection	Corticosteroids: Powerful and long-lasting anti-inflammatory and anti-allergic effect, achieved by inhibiting collagen synthesis, proliferation, and biosynthesis of fibroblasts, suppressing pro-inflammatory mediators, and inducing apoptosis. Therefore, induces the cicatrix to flatten, soften and mature. Options are hydrocortisone acetate, methylprednisolone, dexamethasone, and triamcinolone acetonide. 5–Fluorouracil: Inhibit the synthesis of deoxyribonucleic acid by inhibiting thymine synthase, thus inhibiting the excessive growth of fibroblasts and TGF- β inducing collagen type I expression, inducing lesion flattening. Botulinum toxin type A: Act on cholinergic nerve endings. It is suggested that it may directly regulate the activity of fibroblasts by pausing the fibroblast cell cycle in a non – proliferative state and influencing TGF- β 1 expression. Verapamil: Can reduce the production of extracellular matrix, induce the synthesis of procollagenase in fibroblasts and inhibit the proliferation of IL-6, VEGF, and fibroblasts. Furthermore, it could inhibit the proliferation of fibroblasts and the expression of TGF- β 1 and induce apoptosis of fibroblasts.
Cryotherapy	It would produce tissue necrosis induced by vascular damage secondary to frostbite, which would induce flattening of the lesion.
Laser therapy	Disrupt the high blood flow, decreasing fibroblast proliferation, type III collagen deposition and inflammatory cytokines to reach hypertrophic scars, thereby suppressing their development.
Surgical treatment	Achieves lesion excision and Z or W plasty, designed to change the direction of the scar with a tension – free wound closure.
Post – surgery Radiotherapy	It would produce anti-angiogenesis and successive anti-fibroblast activity inducing apoptosis, resulting in a decrease in collagen synthesis, decreasing the development of keloids.

TGF-β: transforming growth factor beta; IL-6: interleukin-6; VEGF: Vascular Endothelial Growth Factor.

CONCLUSIONS

HS are relevant dermatological entities that can cause pain as well as functional and aesthetic problems, causing significant disruption in daily and long-term activities. It is considered a PHS when HS is accompanied by pain, which is characterized as neuropathic pain resembling a PTTN. In addition, treatment can be challenging due to the unique complexity of each case and the multiple options available for its management without currently having a gold standard. In this case report, the combined therapy between infiltrative local anesthetic and corticosteroid infiltration for PHS showed successful results, being a simple, non-surgical, and effective treatment in relieving pain and the aesthetic characteristics of the scar in the short and long term.

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RESUMEN: Las cicatrices hipertróficas dolorosas (CHD) son sobrecrecimientos fibrosos benignos de tejido cicatricial que manifiestan una sintomatología subjetiva con una fisiopatología poco conocida y la ausencia de un tratamiento estándar, representando un condición desafiante que puede afectar dramáticamente la calidad de vida del paciente. Reporte de Caso: Paciente de 16 años acude con un dolor de intensidad 10 en la escala visual análoga, secundario a una CHD en la región submandibular. El tratamiento se realizó mediante tres sesiones de infiltración de anestésico local peri y sublesional combinado con la posterior infiltración de corticoesteroides. Después de 7 meses la paciente fue dada de alta, retomando todas sus actividades diarias y sin recurrencia del dolor luego de un año de seguimiento. Las CHD poseen características de dolor neuropático y pueden parecerse a una neuropatía post-traumática. Considerando la complejidad única de cada caso y las múltiples opciones disponibles para su manejo, este caso expone un tratamiento efectivo, simple y no quirúrgico que muestra resultados exitosos.

PALABRAS CLAVE: dolor facial, cicatriz hipertrófica, cicatriz hipertrófica dolorosa, lesiones del nervio trigémino, cicatriz, neuropatia posttraumatica, reporte de caso.

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