Sympathetically Maintained Pain Presenting First as Temporomandibular Disorder, then as Parotid Dysfunction

Subha Giri, BDS, MS; Donald Nixdorf, DDS, MS

ABSTRACT

Complex regional pain syndrome (CRPS) is a chronic condition characterized by intense pain, swelling, redness, hypersensitivity and additional sudomotor effects. In all 13 cases of CRPS in the head and neck region reported in the literature, nerve injury was identified as the etiology for pain initiation. In this article, we present the case of a 30-year-old female patient with sympathetically maintained pain without apparent nerve injury. Her main symptoms — left-side preauricular pain and inability to open her mouth wide — mimicked temporomandibular joint arthralgia and myofascial pain of the masticatory muscles. Later, symptoms of intermittent preauricular pain and swelling developed, along with hyposalivation, which mimicked parotitis. After an extensive diagnostic process, no definitive underlying pathology could be identified and a diagnosis of neuropathic pain with a prominent sympathetic component was made. Two years after the onset of symptoms and initiation of care, treatment with repeated stellate ganglion blocks and enteral clonidine pharmacotherapy provided adequate pain relief.

MeSH Key Words: complex regional pain syndrome; pain, intractable; parotitis; temporomandibular joint disorders

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occurrence of sympathetically maintained pain in the head and neck region, but without nerve injury as a clear initiating factor.

Case Report

A 30-year-old woman initially presented with left-side jaw pain and inability to open her mouth wide. She was not aware of any initiating factors and reported that she had experienced this type of pain intermittently since she was 19 years old. The pain had presenting symptoms typical of a left-side temporomandibular disorder (TMD): dull intermittent preauricular pain of moderate intensity that caused sleep disruption, was aggravated by jaw activity and was alleviated by analgesic use. She reported an episode of pain in the left side of her face one year before the onset of the current complaint; that pain had been diagnosed as an ear infection and was treated with antibiotics and surgical drainage. In the past, she had been prescribed gabapentin and opioid analgesics for her pain. Clinical findings, also typical of TMD, included mandibular range of motion less than 40 mm, absence of joint noises and excursions, and tenderness of the left temporomandibular joint (TMJ) and masseter muscle to palpation. Panoramic and tomographic imaging revealed slight sclerosis of the anterior aspect of the right condyle with mild flattening (Figs. 1 and 2). No sensory abnormalities of the face or jaws were detected from the history or physical examination using sharp–dull discrimination and sensation of touch with a wisp of cotton. Based on this information, a diagnosis of left-side TMJ arthralgia and myofascial pain of the left masseter muscle was made.

Conservative treatment was initiated — self-care, a maxillary flat-plane occlusal splint and pharmacotherapy with nonsteroidal anti-inflammatory drugs (diclofenac.

Figure 1: Panoramic radiograph showing no overt odontogenic or osseous pathology.

Figure 2a: Tomography of the right side of the temporomandibular joint indicating mild regressive bony remodelling.

Figure 2b: Tomography of the left side of the temporomandibular joint appearing within normal limits.
and nabumetone) and low-dose nortriptyline — but only minimal beneficial effects were achieved. Lidocaine trigger-point injections on the left masseter resulted in reduction of pain to a minimal level; thus, injections were continued monthly for the following 4 months.

After 9 months of stable pain control, the pain progressively increased in intensity. The patient described it as a pressure sensation in her ear, which she perceived as an ear infection. Swelling and redness was noted in the left preauricular, facial and posterior mandibular areas. Clinical examination revealed that the maximal mandibular opening was less than 30 mm; left-side preauricular areas were tender to palpation although the patient’s left ear tympanic membrane appeared normal. A diagnosis of acute capsulitis with effusion was made and an intra-articular joint injection of corticosteroids was provided. At this time, chronic pain pharmacotherapy was initiated; nortriptyline, titrated to 30 mg before bedtime, resulted in excellent pain relief. Pain, swelling and redness continued to recur intermittently for about one month.

Six months following the TMJ injection, the patient was referred for an oral medicine consultation to assess for parotitis, despite the normal clear appearance of her saliva. A parotid sialogram revealed no calcification or fibrous tissue within the left parotid gland. She was referred to an ear, nose and throat surgeon for biopsy to confirm a tentative diagnosis of Sjögren’s syndrome. Minor salivary gland biopsies of the lower lip, at 2 different times, revealed normal salivary gland histology. Another flare-up of symptoms occurred with the additional new symptom of reduced amounts of saliva. A diagnosis of medication-induced xerostomia with probable secondary infectious sialadenitis was made. A second sialogram was obtained and, again, no abnormalities were detected (Fig. 3). Regional magnetic resonance imaging (MRI), from above the TMJs to below the clavicles, was also normal.

A diagnosis of probable sympathetically maintained pain was made based on the presence of fluctuating autonomic symptoms and the absence of any other organic causes of this patient’s pain complaint. A left-side diagnostic stellate ganglion block was performed using 0.5% bupivacaine. Pre-injection pain intensity was 6/10 on a numeric rating scale. Immediate exacerbation of her left-side jaw and neck pain with final needle placement (which was rated as 10/10) occurred with the pain level reduced to 2/10 15 minutes post-injection. An ipsilateral Horner’s syndrome was present confirming adequate blockade.

Because of the patient’s desire not to receive further injections, pharmacotherapy treatment was initiated, with enteral clonidine starting at 0.1 mg/day at bedtime and increased when sedation was unacceptable to 0.9 mg/day. Topical clonidine was used in a 50% dimethyl-sulfoxide cream, along with gabapentin at 400 mg 4 times a day and Senokot-S (Purdue Pharma, Pickering, Ont.) to counteract the anticholinergic effects of the medication. The pain decreased to an intensity level of 4/10, but over the following 5 months the patient’s symptoms slowly progressed with increasing pain and further spread into her left neck, shoulder and forearm.

During this time, the patient displayed symptoms of depression and in a desperate attempt to alleviate her pain she self-administered cannabinoids. When discussing this, she reluctantly revealed that no appreciable pain reduction was experienced, but smoking did provide a “vacation from her situation.” Consultation with a psychologist was insisted on (previous attempts to refer her to a psychologist had failed).

The idea of a series of stellate ganglion blocks was reinitiated and executed, resulting in excellent pain relief; a pain level of 0/10 was achieved with occasional exacerbations to 2/10. Pain control was so successful that the dose of enteral clonidine was reduced to 0.1 mg every morning and 0.3 mg at bedtime, and the topical cream was discontinued. After 4 months of good pain control with weekly to biweekly stellate ganglion blocks, the patient was lost to follow-up due to relocation of the care provider.

Discussion

Very few cases of CRPS in the head and neck region have been reported in the literature. All can be traced back to identifiable etiologic factors involving trauma — CRPS due to penetrating trauma, surgical procedures in the face and jaw, vascular surgery of the neck, motor vehicle accident leading to head and neck injury, and dental extractions. Our case report is a rare occurrence of sympathetically maintained pain in the head and neck without identifiable nerve injury as a causative factor. Therefore, the most appropriate diagnosis would be CRPS-type I.

The pathophysiology underlying CRPS is not well understood, but small-fibre degeneration has been shown in patients diagnosed with CRPS-type I despite a lack of clinical findings indicating nerve injury. Obtaining
Pain
Dr. Donald Nixdorf, Division of TMJ and Orofacial

also an assistant professor in the department of

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Changes similar to these may

The uniqueness of the current case lies in its symp-

tomatic presentation as TMD pain, otherwise known as

The inability to identify a direct trauma or nerve injury excludes the possibility of

causalgia or CRPS-type II. At the same time, it supports

the diagnosis of TMD, which often presents with un-

identifiable etiology. The clinical findings of spontaneous

excruciating pain not proportionate to the extent of the

pathology of the region and the sympathetic features of

swelling, redness and parotid dysfunction also fulfil the

criteria for CRPS. Further support may be drawn from

the negative sialograms, which ruled out parotid gland

pathology, MRI for the head and neck region, which ex-

cluded vascular pathologies, and the effectiveness of the

stellate ganglion blocks that reinforced evidence for the

involvement of the sympathetic nervous system.

It could be speculated that the involvement of the sympathetic nervous system in the current case resulted from the neuropathic effects of longstanding TMD pain that transformed over time into sympathetically medi-

ated pain. In an animal study of TMJ inflammation and

pain, changes within the nerves innervating the disc were

shown to be induced. Changes similar to these may

result in the expression of adrenergic receptors in the

peripheral sensory fibres resulting in sympathetic dys-

function. This is evidenced by the effectiveness of initial

TMD care. Also, autonomic features were minimal in

the initial presenting symptoms, but they became more

prominent as time passed, with intermittent swelling and

pain exacerbation spreading into the neck.

It is also possible that the sympathetic involvement pre-existent the labelling of the condition as TMD pain, as evidenced by the patient’s admissions to hospital with complaints of preauricular pain and pharmacotherapy with gabapentin before her initial TMD consultation. Also, pre-existing sympathetically maintained pain may have been precipitated by factors, such as minor periph-

eral inflammation and nerve injury secondary to the

treatment provided, TMD pain itself or the prior inner ear infection. This pre-existing sympathetic component could have been merely masquerading as a TMD pain complaint, then parotid dysfunction, requiring time and adequate stressors to cause an exacerbation of this centrally mediated pain phenomenon. The initial inter-

pretation of periauricular hyperalgiesia with intermittent

swelling as being TMD or parotid gland dysfunction would then be best explained as practitioner bias.

Conclusion
This case report is an example of CRPS-type I with sympathetically maintained pain presenting in the head and neck region that required expertise in both orofacial

pain and oral medicine to arrive at an accurate diag-

nosis. We can extrapolate 2 possible clinical scenarios, which are not mutually exclusive, for how this pain pre-

sented: persistent TMD arthralgia and myalgia resulting

in central changes that ultimately led to sympathetically

maintained pain; and sympathetically maintained pain,

masquerading as TMD pain and parotid gland dysfunction

before becoming readily identifiable.

The absence of any identifiable etiologic factors makes this case unique among reported cases of CRPS in the head and neck. It is important for us, as clinicians, to

be alert and closely observe patients with long-standing

TMD symptoms, so that if neuropathic components of

pain arise, they can be identified and treated.

THE AUTHORS

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Dr. Giri is a TMJ specialist with the Minnesota Head and Neck Pain Clinic in Minnesota, Minneapolis.

Dr. Nixdorf is an assistant professor in the department of diagnostic and biological sciences, University of Minnesota, Minneapolis, Minnesota.

Correspondence to: Dr. Donald Nixdorf, Division of TMJ and Orofacial Pain, School of Dentistry, University of Minnesota, 6-320 Moos Tower, 515 Delaware St, Minneapolis, MN 55455, USA.

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References