

## **Case Report**

### **Reflex Sympathetic Dystrophy Syndrome Complicating the Management of TMJ Symptoms. A Case Report**

M. Jeffrey Morton, D.M.D., Mark L. Pitel, D.M.D.

#### **Clinical Report**

A case report and a review of a rare facial pain syndrome is presented. Reflex sympathetic dystrophy syndrome (RSDS) is a condition that is commonly seen and treated by other health practitioners, but which is rarely seen by dental practitioners. Because many of the signs and symptoms of RSDS are similar to TMJ and other facial pain syndromes, it should be an important consideration in the differential diagnosis and selection of proper treatment.

More widely known as "causalgia," RSDS is a complicated, poorly understood condition that has been known for over 100 years. The goals of this paper are to (1) familiarize the dental profession with the symptoms and the currently accepted terminology to describe the condition, (2) present the current theory of the pathogenesis, and (3) describe a case where post-traumatic TMJ symptoms and their treatment were complicated by an associated case of RSDS involving the face.

It is important to state at the outset that the reporting of a case Where the patient responded poorly to therapy is highly significant because it points out the need for the differentiation of RSDS from other conditions more readily managed by the dentist.

#### **Definition of the Condition**

Reflex sympathetic dystrophy syndrome is an extremely interesting condition that is well known to the medical and podiatric professions, but has only rarely been reported in the dental literature. It is a poorly understood neurogenic pain syndrome that appears to involve a disturbance of the reflex pathway of the sympathetic nervous system following some local injury. The syndrome is characterized by persistent pain, discoloration, increased perspiration and elevated cutaneous temperature of the affected part. In severe cases there may be edema, De-ossification of the underlying bone (osteoporosis), and secondary trophic changes such as atrophy of skin and muscle, and grooving and fissuring of nails.

The pain of RSDS usually spreads far beyond the area of the injury. persists long after the area heals, and is not confined to the distribution of a particular sensory nerve. This has been interpreted as involving a change in the internuncial neuron pool.

The uncertain pathogenesis of RSDS, as well as the variability of the symptoms has led to a large number of descriptions and terms that all refer to the same syndrome. The syndrome was first described by Mitchell et al<sup>1</sup> in -1864 who called the syndrome causalgia. which means "burning pain." Dentists are probably most familiar with this term. It has also been widely known as Sudeck's Atrophy: since about 1900. The currently accepted term. Reflex sympathetic dystrophy. originated with Evans<sup>3</sup> in 1947. causalgia. and Sudeck's atrophy are the three most...commonly used descriptors.

Causalgia has been frequently described in the dental literature. however, the reports of RSDS specifically affecting the trigeminal system are very limited. The majority of case reports center around "causalgic like" symptoms following tooth extraction, pulp extirpation. or toothache of nondental origin. Patients describe the pain as persistent. unremitting and burning in quality. giving it a neurotic quality uncharacteristic of true odontogenic pain. Because the pain may often involve several teeth. attempts at treatment frequently lead to multiple dental extractions or endodontic interventions all to no avail. The RSDS may follow trauma, minor or major oral surgery. or infection so the exact dental relationship may be obscure to both dentist and patient. It is likely that some cases diagnosed as craniomandibular dysfunction. TMJ or myofascial pain dysfunction syndromes, which were refractory to conventional therapy, were in fact RSDS or combinations of diseases.

## **Pathogenesis**

RSDS has been found to be caused most commonly by trauma, infections, and neoplasms.<sup>4</sup> Though the actual mechanism of RSDS is still poorly understood. the best current theory is as follows:

1. Peripheral tissue injury causes damage to Sensory neurons. As a result of nerve damage. a barrage of afferent impulses is generated by the dorsal ganglion and propagated both orthodromically and antidromically.<sup>5</sup>

2. Damage to the sensory neuron disrupts the myelin sheath and induces neural sprout formation. The neural sprouts become sensitized to norepinephrine creating an

artificial synapse or 'short circuit' to nearby efferent sympathetic fibers. Autonomic impulses being conducted along vasomotor, pilomotor, and sudomotor pathways cross over to the damaged afferent nerve and induce a nociceptive response.<sup>6,7</sup>

This would explain why causalgic pain is affected by temperature changes and emotional stimulation. It also explains why the most successful therapies to date have been stellate ganglion blockade, adrenergic blockade, guanethidine blockade, sympathetic blocks and sympathectomy.

The increased flow of afferent impulses orthodromically induces an abnormal state of activity in the internuncial neuron center of the spinal cord, this has been confirmed by the work of Erlanger and Gasser.<sup>8</sup>

Augmentation of afferent impulses by the internuncial pool explains why the pain of RSDS is rarely confined to the distribution of a particular sensory nerve.

## **Case Discussion**

The patient, a 20-year-old white female, was referred for TMJ evaluation and treatment approximately one month following trauma received in an automobile accident. During the accident, the patient was thrown forward and apparently struck the right side of her face on the front windshield. The chief complaint was severe right-sided facial pain, limitation of opening, jaw sounds, moderate trismus, and pain during mastication. The pain was described as continuous and throbbing. It was reported to have begun almost immediately following the trauma, was always localized to the right facial region, and was getting worse. In the morning her face felt tight. She noted having her teeth often clenched. She related having the sensation of her face feeling swollen and looking swollen on the right side. The patient also reported unusual cutaneous thermal sensations ranging from cold to hot.

A review of the patient's medical history revealed no significant systemic diseases or allergies. She was hospitalized three times during the year for surgery, including tonsillectomy, cholecystectomy, and a sympathectomy for persistent left-sided rib pain. In the case of the latter surgery, a diagnosis of reflex sympathetic dystrophy had been made and the sympathectomy apparently proved to be successful in eliminating the rib pain. This pain was the result of injury from a previous car accident.

The patient was taking the following medications: Tolectin, Fiexoril, and Talwin. Clinical examination revealed a well-developed 20-year-old female in acute distress

from facial pain. She had a distinctive asymmetry to the face. The right side appeared slightly swollen from the zygoma to the inferior border of the mandible.

The area had a blotchy mottling with irregular zones of pink, white, and red skin coloration. The parotid gland was palpated and felt larger than its contralateral counterpart. The patient had a maximum interincisal distance of 15 mm when opening her mouth.

Palpation of the muscles of mastication, facial, and neck muscles, TMJ, and skin on the right side of the face produced immediate severe pain to the patient. Early clicking of the right TMJ was detected during mandibular depression. Translation of the right condyle was diminished when compared to the left during mandibular opening. Radiographic studies demonstrated no fractures, dislocations, or other significant findings.

Dental examination showed generalized marginal and papillary gingivitis. Considerable plaque, calculus, and materia alba were present on visible tooth surfaces. Numerous carious teeth were noted. A parafunctional habit at clenching was observed.

Our differential diagnosis included post-traumatic arthropathy of the right TMJ, but clearly this would not account for all of the unusual symptoms seen in this patient. RSDS was also suspected because of her previous history of this condition. Confirmation of the diagnosis of RSDS in the face has been documented by successful relief of pain following sympathectomy or sympathetic blockade (usually of the stellate ganglion)<sup>9,12</sup>

A referral was made to a neurologist who confirmed the RSDS problem in our patient by relieving the pain with sympathectomy. Since the pain relief proved only transient, we began conventional supportive TMJ treatment in hopes that minimal disarticulation of the dentition would provide some bruxism relief and patient comfort while the neurologists' comprehensive therapy would be successful in managing the patient's pain.

## **Treatment**

A mandibular acrylic bruxism splint was initially used to disarticulate the tooth-to-tooth contacts and to control the parafunctional clenching habit. Over a five-week period, the mandibular position was gradually shifted from habitual centric position to a protrusive relation. This did apparently reduce some of the TMJ component of the pain, but the overall relief was minimal.

We next initiated a series of needling visits both for localization of any pain trigger zones and for pain control. Initially we used sterile saline, and 3% Mepivacaine (plain) to first localize trigger point locations. Some success was achieved in mapping three locations as shown in **Figure 1**. After providing some pain relief to the patient, the decision was made to try to give some hours of pain relief using the non-vasoconstriction anesthetic and later 2% Xylocaine with 1:100,000 epinephrine and Marcaine with the hope of prolonging the pain relief. It was discussed with the other therapists and the patient, properly noted and of some concern that the later solutions had greater myotoxicity. Hot compresses to increase blood flow, vapocoolant cryotherapy, and conventional TENS treatments were all used with only marginal effect.

Restoration of the patient's carious teeth and periodontal management were completed in a single visit under general anesthesia. This had no effect on the pain. We also tried a variant modality of TENS called the "pain suppressor."<sup>13</sup> which is reported to be somewhat different from the conventional TENS units.

Finally, having reduced the bruxism component of the pain and achieved all that we could within the scope of dental practice, the patient was referred back to the neurologist for a more definitive management of the RSDS problem. Future follow-up indicated poor pain control was achieved through neurosurgical attempts and medical treatment was going toward a more chronic pain management direction.

Discussions of medication use for pain management was being discussed at the time of submission of this paper.

## **Conclusion**

A case is presented where an unusual pain syndrome, Reflex sympathetic dystrophy complicated the diagnosis and management of post-traumatic TMJ symptoms. Diagnosis was based on the history, clinical examination, signs, and symptoms. Confirmation of RSDS was made by relief of pain following sympathectomy. Numerous other therapies were attempted to comprehensively manage the patient's pain. These included bruxism splint, needling of pain trigger zones, moist heat, vapocoolant cryotherapy, TENS, "pain suppressor-TENS" therapy, and restoration of the carious teeth. None could be considered completely successful in eliminating or

controlling significant aspects of the pain. The needling visits were useful in demonstrating and outlining specific trigger zones for the pain. These are outlined in **Figure 1**. Initially, the patient reported high levels of pain relief from the local anesthetic injections for periods of up to three hours; however, she quickly became refractory to these. Some pain relief has come from use of the "pain suppressor." As has been pointed out, the relief of pain by sympathetic blockade or sympathectomy is diagnostic for RSDS. In many cases these are also therapeutic as they were when this patient suffered from RSDS of the back. In this case, however, they produced only transient relief of the facial pain. This may be due to the greater abundance of nerve fibers found in the face and head than other locations often reported for RSDS such as arms and legs. In any event, this case identifies a rare condition that dentists should be aware of and consider in the differential diagnosis of pain syndromes that affect the face and oral cavity.

Further information about RSDS can be obtained by contacting the RSDS Association, P.O. Box 821, Haddonfield, New Jersey 08033.

Reprint request to:  
M. Jeffrey Morton, D.M.D.  
520 Stokes Rd. Rt 541  
Medford, New Jersey 08055

## References

1. Mitchell SW, Moorehouse GR, Keen WW, Gunshot Wounds and Other Injuries of Nerves. Philadelphia. JB Lippincott, Co 1864.
2. Sudeck P: Uber die akute entzundlicke knockentrophie. Arch F Klin Chin 1900 62: 147
3. Evans JA, Reflex sympathetic dystrophy. Report on 57 cases Ann Intern Med 1947; 26: 417
4. DeLorimer AA, Minear WL, Boyd HR: Reflex Lyperemic deossification regional to joints of the extremities. Radiology 1946 40: 227
5. Watt PD, Devor NI. Sensory afferent impulse. originate from dorsal root ganglia as well as from the periphery in normal and nerve injured rats. Pain 1983 17:321-339

6. Bell WE: Orofacial Pain;. Classification Diagnosis. Management. P 303. Third Edition. Chicago. Yearbook Medical Publishers. 1985
  7. Melament IB. Click JR: Sudecks Atrophy. the Clinical Syndrome. J. Am Pad Assoc 1983 73 (7): 362-367
  8. Erlanger I, Gasser HS: Electrical signs of nervous activity. Philadelphia University of Pennsylvania Press. 1937
  9. Jaeger B. Singer F. Kroening R: Reflex sympathetic dystrophy or the face. Report of two cases and a review literature. Arch Neurol 1986 43 (17): 693-695
  10. Khoury R. Kennedy SF. MacNamara TE. Facial causalgia. Report of cases. J Oral Surg 1980 38:782-783
  11. Hanowell ST. Kennedy SF: Phantom tongue pain and causalgia: Case presentation and treatment. Anesth Analg 1979 58: 436-438
  12. Bingham JAW: Causalgia of the face: Two cases successfully treated by sympathectomy. Br Med J 1947 1: 804-805
  - 13 Pain Suppression Labs. Inc. 550 River Dr. Elmwood Park. New Jersey 07407-0441
- 

### **Mark L. Pitel, D.M.D.**

Dr. Mark L. Pitel graduated from the Temple University School of Dentistry in 1982. He is currently in private general practice in Swedesboro. New Jersey. Dr. Pitel serves as vice president of the New Jersey Academy of General Dentistry and chairman of the Speaker's Bureau of the Southern Dental Society. He is a member of the American Dental Association and Academy of General Dentistry-

### **Jeffrey Norton, D.M.D.**

Dr. Jeffrey Morton received his D.M.D. from the University of Pennsylvania School of Dentistry. He is presently in private practice in Medford. New Jersey. Dr. Morton's professional memberships include the American Association or Periodontology. the

Philadelphia Society of Periodontology. the Southern Dental Association. and the  
American Dental Association.

July 1989. Vol. 7. NO. 3 THE JOURNAL OF CRANIOMANDIBULAR PRACTICE  
234