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# Segmental Reflex Sympathetic Dystrophy Involving the Thumb: A Rare Complication of a Varicella Zoster Infection

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The reflex sympathetic dystrophy syndrome (RSDS) as a complication of varicella zoster was first described by Sudek in 1901.<sup>1</sup> In the past 90 yr a small number of reports have associated a varicella zoster infection as the precipitating factor in RSDS.<sup>2–5</sup>

The following case report describes a rare manifestation of segmental RSDS associated with a varicella zoster infection. To the author's knowledge, this is the first published paper of a segmental RSDS precipitated by varicella zoster infection.

## CASE REPORT

The patient was an 84-year-old white woman who was referred to the pain clinic because of persistent burning pain in the left hand and forearm.

Approximately 5 months previously she had been treated for typical symptoms of varicella zoster involving C6, C7, and C8, on the left. The vesicles were noted along distribution of the left radial nerve of the arm and hand. She also had burning pain over the dorsum of the hand, lower arm, and upper arm in the area supplied by the radial nerve. Some pain was located in the nail bed of the thumb. Oral acyclovir, 800 mg, five times daily was prescribed for 7 days.

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During the subsequent 3 weeks the vesicles and rash in the upper arm, forearm, dorsum of the hand, and the first three digits subsided without complicating bacterial infection. She continued to complain of mild burning pain and hyperalgesia in the sites of the healing lesions.

At this time, the patient's attention was focused on the insidious onset of an intense, burning pain in the left thumb. The persistent, progressively severe pain required hydrocodone for relief. Initially the pain affected the phalangeal and metacarpophalangeal joints. The periarticular pain was associated with swelling, redness, and stiffness in the joints. Function and mobility of the thumb became progressively limited. Concomitantly edema, cold and mechanical allodynia, and a cyanotic flush appeared. These symptoms affected the thumb to the level of the metacarpophalangeal joint on the dorsal surface and most of the thenar surface. She could not tolerate a loose-fitting glove because of the allodynia. A cold wind or immersion of the hand in cold water exacerbated the burning pain in the thumb.

The initial examination revealed an alert but depressed elderly woman in severe pain. She held her left arm in her right hand avoiding any contact with the examination table. She also complained bitterly of the intense, burning pain in the left thumb and resisted any passive movement of the hand. There was slight edema of the forefinger, however, the periarticular areas were not tender and mobility appeared normal. The third, fourth, and fifth fingers were mobile and had normal grip strength compared to the right hand. The thumb nail was large, thickened, cracked, and had irregular longitudinal ridges. She was unable to trim the nail because of the intense pain precipitated by the attempt. The skin of the thumb was thin, warm, moist, cyanotic, edematous, and smooth. An alcohol sponge placed on the thumb caused exacerbation of the pain. Light touch of the entire thumb surface produced intense pain. She could be induced to flex the thumb about 1 cm with considerable pain.

An x-ray film of the left hand revealed irregular periarticular patchy areas of osteoporosis and resorption of trabecular bone in the metaphysis of the proximal and distal phalanx of the thumb. The radiologist noted other areas of minimal osteoporosis expected at her age; however, the marked changes of the thumb bones "were consistent with a diagnosis of RSDS."

Thermography studies confirmed the vasodilation in the skin of the thumb. In general, the temperatures of the skin on the left hand ranged from  $30.2^{\circ}$  C to  $32.0^{\circ}$  C in contrast to the range of  $28.4-29.3^{\circ}$  C on the right.

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A series of left stellate ganglion blocks 7 days apart was started. The anterior paratracheal approach was used. Each injection contained 6 ml 2% lidocaine and 4 ml 0.25% bupivacaine. A Horner's syndrome was obtained 5 min after the block was completed. The temperature changes were monitored with the color-coded liquid crystal adhesive patches. These were placed on the thumb, forearm, and upper arm.

Fifteen minutes after the first block, the patient could tolerate passive flexion and extension of the thumb. The burning pain in the left thumb subsided. After 30 min, she was able to actively oppose the thumb, the second, third, fourth, and fifth fingers. The opposed thumb and each finger could firmly grasp a sheet of paper and resist a vigorous attempt to remove it from her grip. The temperature change in the left hand, forearm, and arm increased an average of 4° C. No change was detected on the right. She easily tolerated a cold alcohol sponge rubbed over the surface of her left thumb. One hour after the first block, the assisting nurse was able to trim the oversize thumb nail. The patient was free of pain during the procedure. During the ensuing 7 days, a gradual regression to the condition prior to the block was observed. However, the pain, edema, and cold allodynia were less severe. Despite the recurrence of symptoms, she requested that the stellate block be repeated.

Following each subsequent block a new plateau of stable, permanent improvement was reached. Thus, the anticipated cumulative beneficial effects prompted the author to continue the blocks each week. After the fourth block, the edema, periarticular tenderness, and joint stiffness were permanently eliminated. However, a residual of mild burning pain and cold allodynia persisted in the affected thumb.

The improvement continued after the fifth stellate ganglion block. Mobility and hand grip were equal to that of the right. The edema, allodynia, and periarticular tenderness subsided. The skin color was a slight pink and similar to the color of the other fingers. Thermography indicated temperature values similar to those of the right hand. Gloves were worn without discomfort.

Paradoxically, however, she continued to complain of some mild burning pains of the forearm and upper arm in the radial nerve distribution. She did not, however, complain of any disability and was able to cook and perform other household tasks. It was assumed that she was experiencing mild postherpetic neuralgia in the left arm. Analgesics, TENS, antidepressant medication, and physical therapy were prescribed. This pain abated after 4 months. She terminated all therapy, including TENS, physical therapy, and analgesic drugs. She continued to communicate by phone, but did not desire to return to the clinic.

### DISCUSSION

This patient demonstrated many of the classic findings of stage 2 RSDS in the left thumb. The stage 2 clinical findings include periarticular tenderness, burning diffuse pain, and vasodilation or vasoconstriction. In addition, dystrophic nail and skin changes occur. Patchy osteoporosis, cold and mechanical allodynia, non-pitting edema, joint stiffness, and immobility of the extremity have been reported. Kozin<sup>6,7</sup> and Smith<sup>8</sup> indicate that the manifestations of the second stage may be variable and not be obvious in all RSDS patients.

Our patient demonstrated the radiographic findings of osteopenia, dystrophic skin, nail changes, and vasomotor instability. In addition, the periarticular tenderness, stiffness, and mechanical and cold allodynia findings confirmed the diagnosis. The dramatic reversal of pain, stiff-

ness, and dystrophic and vasomotor changes following the stellate ganglion blocks were additional evidence of RSDS diagnosis.

The majority of the reported cases of RSDS following a varicella zoster infection were of the "glove" or "stocking" distribution in the affected extremity. Several dermatomes were involved with the changes of RSDS. The patients described were in marked contrast to our patient, who had symptoms and findings in the distribution of the superficial terminal branch and the dorsal digital nerves of the left radial nerve innervating the left thumb.

The recent literature contains several descriptions of the segmental manifestations of RSDS. These include a documented case of RSDS limited to the fifth finger<sup>9</sup> and two cases affecting the fourth and fifth fingers. <sup>10</sup> A similar change was described after a nail biopsy. <sup>11</sup> The patella <sup>12</sup> and penis <sup>13</sup> have demonstrated segmental RSDS changes following surgery.

The pathogenesis and pathophysiology of RSDS and sympathetically maintained pain (SMP) are still hypothetical. A plausible hypothesis has been proposed by Roberts. He assumes that a "chain of events" begins with tissue trauma that stimulates unmyelinated nociceptors. The dermal trauma in varicella zoster includes vesicles that may progress to ulcer formation with secondary infection. In our patient, the formation of a vesicle under the thumb nail produced considerable pain and could have easily functioned as a trigger precipitating RSDS or SMP.

The increased nociceptor activity, according to the Roberts hypothesis, <sup>14</sup> excites wide-dynamic-range neurons in the cord. The increased activation of the wide-dynamic-range neurons in turn increases sensitivity to all afferent inputs. This includes the low-threshold mechanoreceptors (A-fiber) and afferent cold fibers that precipitate the mechanical and cold allodynia.

Campbell *et al.*<sup>15</sup> emphasized the role of the sympathetic discharge in the pain and hyperalgesia of RSDS or SMP patients. He postulated that in soft tissue or nerve injury alpha adrenergic receptors "become expressed on nociceptors." The stimuli that produce pain mediated by the low-threshold mechanoreceptors activity increases sympathetic efferent discharge. This generates a release of norepinephrine, which "activates the alpha-adrenergic receptor" on the nociceptors. Thus, increased sympathetic activity leads to low-threshold nociceptor activation, producing pain, which in turn causes a further increased level of sympathetic tone. The sympathetic blockade that occurs as a result of a stellate ganglion block terminates the vicious cycle of pain and increased sympathetic efferent activity.

The complex symptomatology in our patient resulted from the RSDS in addition to postherpetic neuralgia. When the RSDS symptoms were eliminated with the stellate ganglion blocks, the residual postherpetic neuralgia became more prominent and persisted for several months.

In conclusion, a case of segmental RSDS, occurring concomitantly with postherpetic neuralgia, is presented. Recognition of this possibility may initiate early RSDS therapy, thus preventing irreversible dystrophic complications.

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# Delayed Pneumothorax Following Difficult Tracheal Intubation

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Tracheal intubation is associated with a number of complications. <sup>1-4</sup> Most complications are immediately apparent and, once recognized, can be readily treated. Less commonly, problems associated with intubation become apparent at a time distant to the intubation itself. We present a case in which a difficult tracheal intubation led to pneumothorax, pneumomediastinum, and subcutaneous emphysema developing in the postoperative period, after the patient's trachea was extubated.

# CASE REPORT

A 64-yr-old, 47-kg healthy woman with degenerative joint disease was scheduled for a right total knee arthroplasty. The patient's past

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medical history was otherwise unremarkable. She smoked in the past, but stopped smoking 15 yr prior to this admission. Her only previous surgical procedure was a tonsillectomy at 3 yr of age. The physical examination was normal, except for the patient's airway. She had a slightly reduced ability to extend her neck and the distance between the hyoid bone and mental protuberance measured only two finger breadths (approximately 3.5 cm). Otherwise her head and neck were normal in appearance with normal dentition and full range of motion at the temporomandibular joint. Despite these abnormalities, examination of the patient's oropharynx provided good visualization of the soft palate and uvula, consistent with a class 1 airway as described by Mallampati  $et\ al.^5$  Preoperative chest x-ray and laboratory test results were normal.

Anesthesia was induced with 250 mg sodium thiopental and 10 mg morphine sulfate. Ease of assisted ventilation was confirmed and 5 mg vecuronium was administered to facilitate tracheal intubation. After achieving adequate muscle relaxation as documented by a peripheral nerve stimulator, laryngoscopy was performed using a Macintosh #3 blade. The larynx could not be visualized. Assisted ventilation of the lungs via face mask was reinstituted, head position altered, and a second attempt at laryngoscopy made without improvement of vocal cord visualization. A subsequent attempt was made to visualize the vocal cords by a second anesthesiologist. In addition to manipulation of the patient's head, gentle posterior pressure was applied to the cricoid cartilage. This facilitated visualization of the posterior aspect of the larynx, although the vocal cords still could not be visualized. Tracheal intubation was accomplished using a 7.0-mm endotracheal tube (ETT) with a stylet, which was used to provide steep anterior angulation of the tip