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Reflex Sympathetic Dystrophy Syndrome in Pregnancy

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The history commonly associated with reflex sympathetic dystrophy syndrome (RSDS) includes blunt or penetrating nerve trunk or plexus trauma. However, RSDS has appeared following uneventful operative procedures, and has even developed spontaneously.¹⁻³§ There are no previous reports of RSDS presenting secondary to carpal tunnel entrapment during pregnancy. Management with a series of unilateral stellate ganglion blocks restored the patient's arm and hand function for the remainder of the pregnancy and postpartum period.

REPORT OF A CASE

A 38-yr-old, gravida 3, para 1 woman, at 31 weeks gestation, had severe burning and aching pain in both hands, right greater than left, of 6-8 weeks' duration. Itching and redness of her hands were first noted 3 weeks prior to the onset of pain. By neurologic examination, edema, hyperhidrosis, and severe dysesthesia in her hands were evident. The patient postured to prevent movement of, or contact with, the skin of her right hand. Her right hand pain and hypersensitivity were so severe that it prevented close sensory or motor examination of the extremity, and precluded any electrodiagnostic studies of the right arm. A nerve conduction study of the left upper extremity was obtained, however, which suggested median nerve entrapment at the wrist (*i.e.*, carpal tunnel syndrome) and ulnar nerve entrapment-compression at the left elbow. Electromyographic studies confirmed the relatively severe entrapment neuropathies of the left side.

On our evaluation, she related symptoms progressing in severity since onset, severely limiting use of her left arm and preventing use of her right arm. The patient scored her right and left hand to be a ten and nine, respectively, on a 10 cm linear visual-analogue pain scale. Laboratory studies revealed normal hemoglobin and red blood cell indices. The erythrocyte sedimentation rate was 40 s. The coagulation profile was within normal limits. Assays for folate and vitamin B₁₂ were within normal limits. The rapid plasma reagin (RPR) was negative, rheumatoid factor was positive, and antinuclear antibody (ANA) was less than 1:40 dilution. Thyroid function tests, serum electrolytes, blood urea nitrogen, and serum creatinine were all normal.

After obtaining informed consent, the patient was placed supine with uterine displacement assured. A right stellate ganglion block with

10 ml of 1% lidocaine was performed. Increased skin temperature, improved venous architecture, and a right Horner's syndrome concomitant with complete relief of pain in the right hand were observed within 2 min. Left hand symptoms were also completely relieved within 10 min. Pain-free, the patient demonstrated the bilateral use of her hands and digits for the first time in several weeks. Sensation to pin prick was well maintained when tested over the lower cervical and upper thoracic dermatomes on the right and left sides. Fingertip numbness to pin prick was demonstrated bilaterally at this time distal to the wrist in all but the fifth digits. The patient remained pain-free for the next 12 h, after which the pain returned in only her right hand, although less severe (a pain score of 8). On day 2, the patient was then treated for her pain by the orthopedic surgery service with bilateral carpal tunnel steroid injections and wrist splints. Within hours, the painful ache and skin sensitivity were absent, and only "stiffness" remained. Unfortunately, 24 h later on day 3, the pain, ache, and dysesthesia returned to the right hand only (a pain score of 5). On day 4, a second right stellate ganglion block with 10 ml of 1% lidocaine completely relieved her symptoms. The patient was discharged from the hospital and subsequently followed in our outpatient facility 2 days later. Her right hand discomfort had returned, although markedly less severe (a pain score of 5) and limited only to her right thumb and index finger. A third right stellate ganglion block again completely relieved her pain, this time for 4 days. Now only residual "soreness" (a pain score of 3) in her right index finger and thumb remained. She was treated with two subsequent right stellate ganglion blocks with progressively prolonged pain-free intervals (6 days to 2 weeks). Three weeks later, she reported a relapse of symptoms in her left hand that she attributed to excessive use. A sixth right stellate ganglion block gave complete bilateral pain relief for 2 weeks. At 37 weeks gestation, she was admitted to the labor and delivery suite in active labor.

An intrapartum diagnosis of fetal distress necessitated emergency abdominal delivery under general anesthesia of a 37-week-old living male child. The umbilical artery and vein pHs were measured as 7.13 and 7.18, respectively; with Apgar scores of 4 and 9 at 1 and 5 min after birth. The child was discharged after an uneventful nursery stay. Postpartum, the patient has reported no pain, burning, swelling, aching, nor dysesthesia in either of her hands. She has residual numbness in the tips of the thumb, and the second, third, and fourth fingers bilaterally distal to her wrist. Sixteen weeks postpartum, the patient denies any return of burning and dysesthetic pain, and to date has not exercised a surgical option for her CTS.

DISCUSSION

RSDS is characterized by pain, hyperesthesia, vasomotor disturbances, and dystrophic changes that usually improve with sympathetic denervation.^{4,5} It is associated with a variety of precipitating factors including: soft tissue or bony injury, infection, thrombosis, malignancy, immobilization with a cast or a splint, connective tissue disorders, brain tumors, and even bed rest.¹⁻³§ The symptoms begin gradually, days or weeks after an injury, but may manifest immediately.²§ Our patient first manifested her symptoms in her second trimester. Her initial complaints of aching pain pro-

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gressed to incorporate burning dysesthesias as her primary symptoms.

Carpal tunnel syndrome (CTS) occurring during pregnancy has been described by numerous authors.⁶⁻⁹ Its reported incidence during pregnancy is approximately 25%.⁷ Complaints of tingling and numbness in the fingers are common with sleep disturbances and bilateral involvement present in 75% of cases.⁷ Symptoms of median nerve compression may occur in 41% of patients. Measurement of nerve conduction velocity across the carpal tunnel remains the standard method of verifying a clinical diagnosis of CTS.^{10,11} Electrodiagnostic studies reveal delayed sensory conduction in 18% and delayed motor conduction in 7% of patients.⁷ Management of CTS during pregnancy usually incorporates conservative measures, such as wrist splinting and steroid injection.^{8,9} Vitamin deficiencies, specifically B6, during pregnancy have been implicated as an etiology of CTS.^{11,12} Vitamin supplementation has been successfully used to treat CTS during pregnancy. Our patient received vitamin B complex and multivitamin supplementation during her pregnancy. Surgical decompression of our patient's likely right and left CTS was offered to her; however, she felt that surgical recovery, which would place her wrists in casts or splints, would impair her ability to care for her children and husband. Furthermore, since we could not guarantee that surgery would cure her burning pain, she preferred treatment with stellate ganglion block that had previously relieved her pain. Connective tissue disorders and alcohol abuse were all addressed and dismissed as possible etiologies in the evaluation of our patient.

Of particular note was electrophysiologic evidence of ulnar nerve involvement. Although ulnar nerve involvement has reportedly accompanied CTS in pregnancy, its involvement is not typical, and suggests a more generalized process than that attributed to median nerve compression at the wrist alone.⁷ Although the diagnosis of RSD is primarily a clinical one, the best approach to confirm the presence of RSDS is the use of differential neural blockade.^{4,12} The sensitivity of roentgenographic evaluation in suspected RSDS is low, approximately 39%, and, even when positive, represents advanced trophic changes that are not entirely specific for RSDS. Scintigraphic evaluation is considered more specific for RSDS; however, her pregnancy precluded this type of study.¹⁷

There is little significant direct effect of low doses of lidocaine on the fetus. There are no known effects of stellate ganglion blockade on the viability of pregnancy or uteroplacental blood flow. Stellate ganglion blockade was, therefore, employed to diagnostically confirm and subsequently treat our patient. Beneficial results from serial treatments with stellate ganglion blocks have been

reported in 49-80% of non-pregnant patients with RSDS.¹⁻³ Other treatment modalities include intravenous regional blockade with steroid preparations or with the catecholamine depleting agents guanethidine and reserpine, systemic alpha receptor blockade with the orally administered phenoxybenzamine, and systemic steroids.¹³⁻¹⁷ Although the success rates of such alternative treatment modalities have, in some studies, approached that achieved with serial paravertebral sympathetic blockade, the latter is generally felt to be more effective and, in this patient, exposed the fetus to less systemic maternal medication.

The dramatic and complete response to right stellate ganglion blockade helped confirm the diagnosis and successfully relieved the pain. The extent and severity of the patient's symptoms decreased and the pain-free intervals lengthened with serial treatments. At no time did the patient complain of numbness in her tongue, ringing in her ears, or lightheadedness. She remained oriented and responsive without manifesting any symptoms of systemic absorption of lidocaine. Several minutes after the block, lower cervical and upper thoracic dermatomes remained sensitive to pin prick bilaterally, excluding epidural or subarachnoid spread as a possible explanation for her contralateral relief. Bilateral median nerve distribution numbness, however, distal to the wrist, was discovered after the first right stellate ganglion block. This numbness was very likely present for some time prior to our discovery; however, the hyperesthetic pain in her hands had, up until now, prevented a close sensory examination. After the first right stellate ganglion block, all left-sided symptoms also completely resolved.

Bilateral relief from a unilateral block is not a common phenomena, although bilateral symptoms from a unilateral etiology have been reported by Livingston¹⁸ and Bonica.¹⁹ Accordingly, reflex disturbances in long-established pain syndromes have been known to spread to involve contralateral limbs. This contralateral pain has been described as a mirror image to the more severe initial insult.¹⁸ It is postulated that, in reflex dystrophy, an irritative nerve lesion serves as a focus of chronic irritation, from which an abnormal number of impulses arise and constantly bombard the spinal cord and upset the normal functioning internuncial pool.¹⁹ The abnormal activity of the internuncial pool spreads upward, downward, and across to implicate other neuron systems and also the anterior and lateral motor horn cells, resulting in excessive skeletal and smooth muscle activity.¹⁹ Consequently, there is muscle spasm and vasospasm that produces hypoxemia and metabolites that serve to stimulate abnormally the sensory nerve ending and, in this way, furnish new sources for pain and reflexes.¹⁹ This, in turn, aggravates the central disturbance in the cord, thus setting up a vicious cycle.¹⁹ In

our patient, it is, therefore, possible that the interruption of aberrant reflex activity originating from a unilateral irritative lesion that had recruited internuncial pool neurons could, thus, remove the contralateral symptomatology. This would appear to explain the bilateral relief of symptoms in our patient achieved with a unilateral block. Reflex disturbances in long-established pain syndromes may manifest contralateral symptoms as a mirror image to the original lesion.¹⁸ Our patient had contralateral symptoms, although her pain was not a particularly long-established syndrome. Her relatively recent history no doubt contributed to our successful interruption of aberrant reflex activity that we postulate initiated her contralateral pain in the first place. Postpartum, she has related no complaint of burning dysesthesia, but only median nerve distribution finger tip numbness. Future electrodiagnostic evaluation will determine the need for carpal tunnel decompression.

In summary, we have presented a pregnant woman who spontaneously developed a bilateral reflex sympathetic dystrophy. Diagnostic studies identified median nerve compression and carpal tunnel syndrome. Serial right stellate ganglion blockade provided our patient complete and progressively prolonged relief from her bilateral RSDS, allowing her to experience a more comfortable third trimester and postpartum period.

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Bupivacaine for Caudal Analgesia in Infants and Children: The Optimal Effective Concentration

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Three of the important variables that determine the effectiveness of caudal blocks for postoperative analgesia are: the specific local anesthetic, the volume of solu-

tion, and the concentration of local anesthetic solution. The effects of the first two variables have been studied previously in infants and children receiving caudal blocks.¹⁻⁶ However, the optimal concentration of local

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