Unexplainable Nondermatomal Somatosensory Deficits in Patients with Chronic Nonmalignant Pain in the Context of Litigation/Compensation: a Role for Involvement of Central Factors?

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ABSTRACT. ***Objective***. To address the prevalence and characteristics of nondermatomal somatosensory deficits (NDSD) in subjects with chronic pain in the context of compensation/litigation.

Methods. Data were collected via standardized history, examination, and patient- as well as physician-drawn body maps in a consecutive series of 194 subjects seen for the purpose of an independent medical examination.

Results. Forty-nine subjects (25.3%) with primarily widespread pain (often diagnosed as fibromyalgia) presented with hemisensory or quadrotomal deficits to pinprick and other cutaneous stimuli on the side of lateralized pain or worse pain. The NDSD limbs often had impairment of vibration sense (not infrequently associated with "forehead vibration split"), reduced strength, dexterity or movement, and extreme sensitivity to superficial skin palpation or profound insensitivity to deep pain. Spatial, temporal, qualitative, and evolutionary patterns of NDSD emerged associated with cognitive/affective symptoms. NDSD subjects were more often born outside Canada, more likely to be injured at work, present with abnormal pain behavior, and have negative investigations.

Conclusion. NDSD are a prevalent problem associated with chronic pain. Future research should explore the prevalence of NDSD in other pain populations, the role of personality and related factors, and the underlying biological substrate of these deficits. (J Rheumatol 2001;28:1385–93)

Key Indexing Terms:
CHRONIC PAIN JURISPRUDENCE HYPOESTHESIA

Nonanatomical sensory deficits have been described for hundreds of years. We use the term "nondermatomal somatosensory deficits" (NDSD) for the purposes of this study. A commonly held viewpoint associating these deficits with "psychogenic" or "nonorganic" pain is best expressed by the following statement: "Psychogenic sensory deficits are diagnosed by their variability, sharp midline transition, and non anatomic dermatomal distibution"[1]. In addition, the presence of these NDSD is one of several criteria for the diagnosis of "non organic" pain[2].

Our group has observed NDSD in chronic pain patients and their behavior under the influence of intravenous administration of sodium amytal, a medium action barbiturate[3,4]. Only 2 empirical studies[5,6] have looked at the prevalence of NDSD in chronic pain populations with the specific diagnosis of diffuse myofascial pain or fibromyalgia (FM). In these 2 studies the prevalence of NDSD ranged between 38 and 40%, but specific subgroups seemed to have a much higher prevalence[5]. We investigated the prevalence and characteristics of NDSD in subjects with chronic nonmalignant pain in the context of litigation/compensation.

MATERIALS AND METHODS

The sample studied consisted of all subjects referred to the senior author (AM) for the purposes of an independent medical examination over the course of 3 years (1996–1999). The following were collected at the time of evaluation: (a) standardized demographic data forms; (b) body maps where patients marked their pain areas; (c) standardized history with attention to original and current pain complaints; (d) behavior displayed through the interview and physical examination; (e) thorough neuromusculoskeletal examination; and (f) documentation of gross cutaneous and deep sensory abnormalities by the examiner in body maps identical to those used by the patient. Cutaneous sensation was tested via a soft brush (for light touch), a pinprick wheel for pain, and a cold roller for gross cold perception; and vibration sense was tested by a 128 Hz tuning fork. Pain arising from deep structures (muscles, periosteum, etc.) was tested by manual pressure in all cases and wherever possible by pressure algometry over multiple bone prominences in the upper and lower extremities. The latter method has been described[7]. All previous files including assessments by other physicians or health care professionals, records and radiological/electrophysiological

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investigations, as well as psychological reports when available, were thoroughly reviewed.

Nonparametric statistics were used for analysis. Chi-square (with Yates' continuity correction for 2 × 2 contingency tables) was used for intergroup comparisons; $p < 0.05$ was considered statistically significant. A Bonferroni correction for multiple comparisons was used.

RESULTS

The sample consisted of 194 consecutive subjects seen for an independent medical examination. The female/male ratio for the whole group was 2/1, mean age 44 years, and average duration of pain complaints 5 years. The majority of subjects (62%) were born outside Canada, while the primary cause of disability was a motor vehicle accident (58.2%), irrespective of the referral source. The 2 largest referral sources were the plaintiff's lawyer (primarily in no-fault accidents) and Canada Pension, each accounting for about one-third of referrals.

Of the total number of subjects, 49/194 (25.3%) were found to have NDSD localized at the site of pain or worse pain (if pains involved both sides of the body). The demographic characteristics of both the NDSD and non-NDSD subgroups are summarized in Table 1.

In the NDSD subgroup of 49 subjects, deficits to pinprick were found in a hemisensory distribution in 57%, in a quadrotomal or whole limb distribution in 30.6%, in 3 quarters of the body in 4.1%, and the whole body (sparing the face) in 8.2%. In the latter group of 4 subjects, hypoalgesia involved the whole body below the level of the neck, where pinprick was felt as distinctively dull compared to the sharp/painful prickliness experienced in the face. In general, the sensory deficits were usually much larger than the actual areas of pain marked by subjects in the body maps. While some patients reported awareness of dense or intermittent/recurrent "numbness," others became quite alarmed to discover the deficit during examination. Pinprick abnormalities ranged from mild hypoalgesia (pinprick still felt as prickly but clearly less than a control area), to moderate (prick was felt as dull), to severe hypoalgesia/anesthesia (the subject could feel no stimulus at all with eyes closed). Light touch and cold perception were equally or less obviously suppressed. Quite often, vibration sense was reduced or lost at the site of NDSD, and several patients had a classic "vibration split" at the forehead. Notably, on some occasions the subject could present with some hyperesthetic abnormality to pinprick and other cutaneous modalities, and later on reverse to hypoesthesia.

The NDSD borders were either sharply demarcated or to the contrary, blurred, but were clearly nondermatomal in distribution. Their variability was demonstrated on several occasions when the NDSD would "shrink"or "expand," even in the course of the same examination or during different examinations by the same examiner. However, on

Table 1. Group demographics.

	Whole Group	Non-NDSD Group	NDSD Group
Sex, %			
Female	66.5	70.3	55.1
Male	33.5	29.7	44.9**
Mean age, yrs (range)	44.2 (4–77)	43 (4–75)	47.3 (32–77)
Mean duration of pain complaints, yrs (range)	5 (4 mo–33 yrs)	4.6 (4 mo–19 yrs)	6.3 (3 mo–33 yrs)
Country of birth, %			
Canada	38	43	25
Other	62	57	75*
Cause of pain, %			
MVA	58.2	61.4	49
Spontaneous	16.5	18.6	10.2
Work related	16	11	30.6*
Other	9.3	9	10.2
Referral source, %			
Plaintiff	39.7	43.4	28.6
Lawyer	35.1	32.4	43
CPP	13.4	13.1	14.3
Rehab. Facility	6.7	6.2	8.2
Defense Medical	4.1	4.1	4.1
LTD	1	0.7	2
Other			

* Indicates statistical significance.
** Approached statistical significance.
MVA: motor vehicle accident.
CPP: Canada Pension Plan related payments.
LTD: longterm disability

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other occasions they could be remarkably reproducible and unchanged in size or intensity during repeat examinations or followup assessments. Truncal NDSD borders would stop at the midline of the torso or a few inches off the midline on the ipsilateral or contralateral side. At times, at the beginning of the examination, the subject seemed to perceive some sensory hypoalgesia in a patchy distribution. With repeat pinwheel testing over a lengthy examination, hypoalgesia would become quite noticeable and well definable.

Manual pressure and pressure algometry of the NDSD extremities often revealed significant sensitivity diffusely, or to the contrary, dense insensitivity. Occasionally the anesthetic or hypoesthetic limb could present with significant tenderness to gentle palpation during one examination and reverse to deep pain insensitivity during the next visit or vice versa, while the cutaneous anesthesia/hypoesthesia remained unchanged. These phenomena are illustrated in case 1 reported below.

In terms of NDSD onset (information retrieved upon review of documentation forwarded and descriptions by the patient), most deficits seemed to be of gradual onset as the pain persisted or increased. In several documented cases, however, numbness was of acute onset upon the impact (injury at work or particularly car accident) or was documented in explicit consultation notes as early as a month after the onset.

The phenomenon of expansion and spreading of NDSD is illustrated in case 2. Modification of NDSD was shown to occur under pleasurable activities, distraction, and particularly during the infusion of intravenous sodium amytal (SA). Occasionally, short term, goal directed mobilization with a supportive physiotherapist would produce lasting improvement and rarely reversal of the NDSD.

Some patients seemed more concerned about the presence and spreading of NDSD than the presence of pain. Patients with dense NDSD in their lower extremities had substantial problems driving a car (would not feel the brake, etc) or were falling, finding the numbness equally or more disabling than pain itself. Some patients with dense deficits expressed the feeling that the limb was "alien" or "not theirs." Motor abnormalities (alterations in gait, posture, range of movement, or dexterity) almost always accompanied NDSD and at times they were striking, rarely associated with frank paralysis.

Four patients were ultimately admitted to the inpatient service. In 2 of these patients the sensory findings almost normalized under intravenous SA infusion in tandem with pain relief (phenomenon exemplified in case 1). The 3rd patient, who had a hemisensory deficit and diabetic neuropathy in the distal limbs, failed to alter her deficits under SA infusion despite some pain relief. The 4th refused to cooperate or respond to our sensory and motor testing, therefore we were unable to ascertain what happened to her sensory findings and subjective pain. It is worth reporting that the latter "lost" all her pain in the symptomatic lower extremity when dense hemianesthesia in the same site appeared abruptly prior to her admission to our unit (a phenomenon observed very rarely in this group). Nevertheless, she maintained multiple other pains throughout her body.

Table 2 presents an overview and summary of findings and observations outlining the temporal, spatial, qualitative, and evolutionary characteristics of NDSD, as well as associated cognitive/affective features, inciting events, reported symptoms, and response to interventions. Significant differences emerged between the non-NDSD and NDSD subgroups. (Asterisk below indicates comparisons that were significant even with Bonferroni correction.)

Demographic variables. Those not born in Canada ($p = 0.02$) or injured at work ($p = 0.009$) were more likely to have NDSD. Men had a greater tendency to present with NDSD, but the data did not reach statistical significance ($p = 0.05$).

Pain and sensory abnormalities. NDSD subjects were more likely: (1) To have pain lateralized or worse on one side of the body ($p = 0.0000001$*). However, there was no side prevalence, i.e., more right or left sided abnormalities; (2) To complain of more pain and have increased numbers of pain sites compared to their original complaints ($p = 0.002$ and 0.008, respectively); (3) To present with abnormalities of deep pain perception as measured by algometry or on manual pressure (excessive sensitivity or to the contrary insensitivity within the NDSD area) ($p = 0.000003$*); and (4) To complain of diffuse tenderness over the NDSD regions on skin rolling ($p = 0.005$).

Behavioral and disability variables. NDSD patients were more likely: (1) To present with inability to work ($p = 0.002$), with almost all unemployed versus 31% of non-NDSD subjects who were still employed; (2) To display unusual/bizarre gestures or posturing ($p = 0.0004$*), grimacing ($p = 0.0004$*), or guarding ($p = 0.002$); and (3) To demonstrate significant differences in straight leg raising between distraction (sitting position) and confrontation (direct examination in the supine position) ($p = 0.000003$*). Significant difference had been arbitrarily defined by the examiner to be 30° or more between distraction and confrontation testing.

Overall, however, there were a number of characteristics shared by the 2 subgroups. For example, most subjects in both subgroups suffered from pain in several body regions and had not improved after the original event (even if the NDSD group had actually gotten worse, as reported above). Table 3 shows number and evolution of pain sites over time between the NDSD and non-NDSD subgroups.

CASE REPORTS

Case 1. A 39-year-old woman was seen for independent medical examination 2.5 years after a motor vehicle accident. She was the belted driver of a medium size car that was rear-ended by a jeep. She immediately went

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Table 2. Qualitative characteristics of non-dermatomal somatosensory deficits (NDSD).

Temporal
- Most often develop post-event gradually, in tandem with pain spreading and worsening, but occasionally abruptly at time of inciting event
- At times disappear spontaneously (if pain decreases)

Spatial
- Ipsilateral to the site of lateralized pain or worse pain in case of diffuse symptoms
- Not confining to known peripheral nerve or dermatomal distribution and may involve a whole body quadrant, a part of a limb or half of the body
- The borders could be diffusely or sharply demarcated, while size may remain stable for years or substantially change from time to time
- Usually correspond to much greater body areas than those regions where pain is felt
- Unilateral deficits usually (but not always) do not cross the midline
- May increase in size in tandem with pain increase
- At times pain may be experienced proximally while loss of sensibility is experienced distally

Qualitative
- Primarily involve pinprick but also light touch and cold perception (pinprick hypoalgesia can vary from mild reduction of prickly sensation to complete anesthesia)
- Manual palpation of the NDSD areas may demonstrate (a) intense sensitivity to gentle palpation or to the contrary (b) profound insensitivity (better demonstrated by algometry), at times interchangeable
- Vibration sense is often impaired or absent within the NDSD area, frequently associated with classic "forehead" split
- Motor abnormalities are very frequent in the NDSD limbs, consisting of decreased range of movement, reduced dexterity, abnormal posturing, guarding or gait abnormalities and occasionally paralysis
- Occasionally sudomotor changes, particularly persistent alterations of limb temperature, may be associated with limb NDSD, even in the absence of significant immobility or guarding
- Often, repeat testing enhances the perception and intensity of NDSD

Inciting event
- Original physical injury usually, but not always, insignificant and/or minor
- Often associated with emotional trauma at the time of inciting event

Cognitive/affective
- Most subjects partially aware or usually unaware of NDSD
- Significant mood/anxiety disorder and/or abnormal pain behavior are evident

Reported symptoms
- Diffuse severely burning, or deep aching and "heavy" pain
- Gait and limb use difficulties
- Patients "don't feel the floor", "seem to walk on feathers" or their numb part feels as if they had "an injection of local anesthetic as when they visit the dentist"
- Severe abrupt-onset limb numbness can occur as if the limb is "dead" or feels like "wood"
- Patients often drop things or have loss of leg control and fall due to perceived "weakness" (with or without surge of pain)

Response to interventions
- Often resistant to multiple physical modalities, medications, and/or blocks or procedures
- Occasionally respond to distraction, reduction of distress and/or underlying psychiatric disorder and relaxation techniques with positive reinforcement or goal-directed strengthening and mobilization in conjunction with establishment of good rapport between therapist and patient
- Cutaneous sensibility often normalizes and pain substantially or totally resolves under intravenous infusion of sodium amytal

Table 3. Number and evolution of pain areas over time (numbers represent %).

	NDSD Subgroup	Non-NDSD Subgroup
Pain areas < 3	20.5	30.4
Pain areas > 3	79.5	69.6
Number of pain areas since onset		
Same	31.3%	51.9
Increased	66.7*	43.4
Decreased	—	3.9
Unknown	2.1	0.8

* Statistical significance.

"blank" experiencing "total body pain," being unable to move any limbs. She was taken to the emergency department by ambulance and released the same day. No loss of consciousness was documented and no fractures were found. She underwent several physiotherapy sessions with both passive and active modalities, chiropractic management, aquatherapy, etc. She tried nonsteroidal antiinflammatory drugs, tricyclic and other antidepressants, muscle relaxants, combination analgesics, benzodiazepines, etc., without much relief. Investigations failed to disclose ongoing musculoskeletal or neurological pathology. Nevertheless, she became near house- and bed-bound and even needed help with personal hygiene by an extremely supportive and caring husband. On a body map she indicated near total body pains with clear lateralization to left. She reported numerous physical, systemic, and psychological symptoms. During the interview she presented with flat affect and wept a couple of times. She walked stooped over, drag-

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ging the left leg and holding the left arm immobile close to the chest, with an appearance reminiscent of a patient with stroke. She had obvious signs of a 2 week old burn in her left forearm that she sustained during cooking and did not feel. On examination, she displayed exuberant pain behavior, moaning and groaning, holding on to walls and cupboards, and almost falling over without support. Sensory examination revealed striking and dense anesthesia to pinprick, touch, and cold and complete loss of vibration perception in the left side of the body including left face/forehead, torso, and left limbs. On pressure algometry she was unable to perceive pain at the maximum values of pressure in the left side (22 pounds/cm^2 with a Pain & Diagnostics hand-held algometer). Figure 1 shows her self-drawn pain map and the sensory deficit to pinprick found on examination. Figure 2 shows deep markings from the pressure algometer over the left forearm, which failed to make her perceive pain at the extremes of the available pressure range.

Upon admission to our inpatient unit, her dense cutaneous deficit persisted but she had reversed her deep pain insensitivity to significant sensitivity to gentle manual palpation. This reversal lasted for one day only.

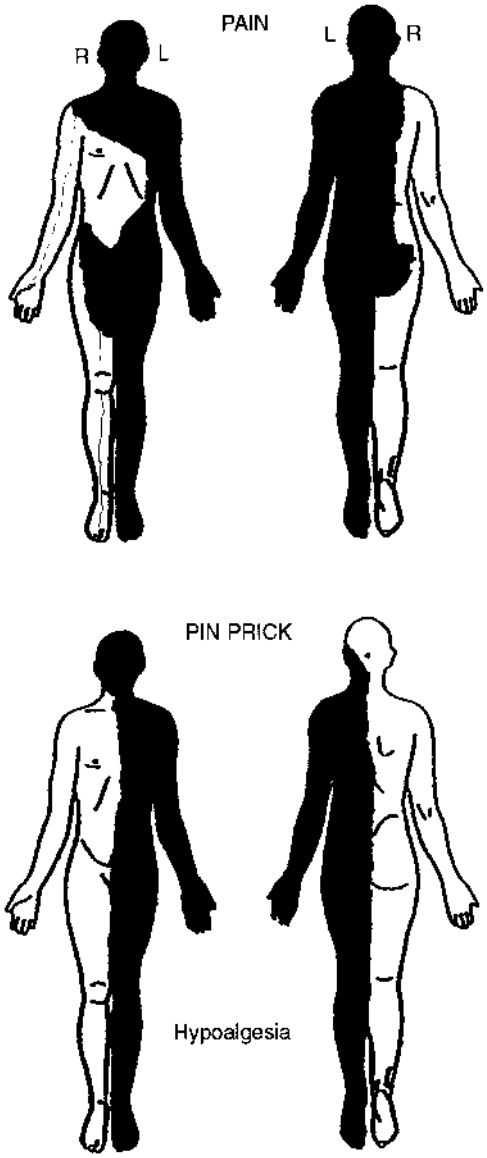

Figure 1. Case 1. Top: patient-drawn pain map. Bottom: area of dense anesthesia to pinprick as detected on sensory examination.

Subsequently, sensory findings nearly returned to normal, with significant pain reduction and amelioration of her behavior during intravenous SA infusion. There were marked elevations of the first 3 MMPI-2 (Minnesota Multiphasic Personality Inventory) clinical scales, suggesting that psychological conflicts were channeled into somatic symptoms and complaints. The team concluded that a very substantial psychological element was contributory to this woman's perceived pain and disability. Cognitive-behavioral treatment together with physical restoration and exploration of family dynamics was recommended.

Case 2. A 45-year-old man was seen for independent medical examination almost 3 years after a minor rear end motor vehicle accident. He was the belted driver of his car. Upon impact he experienced "immediate" neck pain but was able to drive the car home. He saw his general practitioner the same day and subsequently "laid on the floor for 3–4 weeks" with "pain all over." He did not respond to conservative treatments. Multiple radiological and electrophysiological investigations or consultations failed to reveal underlying pathology. Several psychological reports concurred with the DSM-IV diagnosis of pain disorder associated with psychological factors. In addition to severe problems associated with depression and anxiety, certain personality characteristics ("rigidity, tendency to overreact to innocuous events, and use of somatic complaints to avoid facing emotional difficulties") were thought to be contributing to his presentation.

During the original interview he reported headaches, low back pain, and primarily right side body pain. Right leg pain developed 3 months after the accident (associated with episodes of severe acute numbness) and right hand pain one year later. He also reported a host of psychological and constitutional symptoms plus intense posttraumatic stress reaction. He looked despondent with variable gait and variable range of movement in the neck observed under distraction. During the examination he hyperventilated, became flushed, profusely sweating, and presented very limited range of movement in axial and limb examination and multiple signs of inconsistency. An inconsistent and fleeting deficit to pinprick and vibration sense was noted in the right side of the body. He was reassessed 2 years later. At that time he presented with expanded and worsening right side pains and new complaints of severe burning in the arm and leg, as well as right facial numbness. He continued to have the same intense psychological and constitutional problems reported during the first assessment. This time, apart from his intense pain behavior, sensory findings were striking with well defined hemisensory hypoalgesia to pinprick, touch, and cold, and reduction of vibration sense in the right leg. As well, remarkable temperature difference (with the hypoesthetic side colder) was observed, ranging between 1 degree Centigrade for the hand and 3 degrees Centigrade for the leg, despite his using the arm (albeit in a guarded fashion), and ambulating with a limp. Figures 3 and 4 illustrate pain complaints and sensory findings to pinprick during the 2 separate visits.

DISCUSSION

Our study describes the existence of nondermatomal somatosensory deficits covering large parts of the body in 25% of study subjects. These subjects seemed to have little or no detectable physical pathology accounting for their chronic pain — most reported chronic widespread pains, were often diagnosed as suffering from FM or diffuse myofascial pain syndromes, and displayed rather intense pain behavior and disability.

There are certain weaknesses in the study relating to methodological issues: (a) Quantitative analysis was performed only in certain variables that were systematically retrieved, i.e., demographic data, pain and sensory abnormalities, and behavioral and work related variables. Qualitative observations served specifically to give a global

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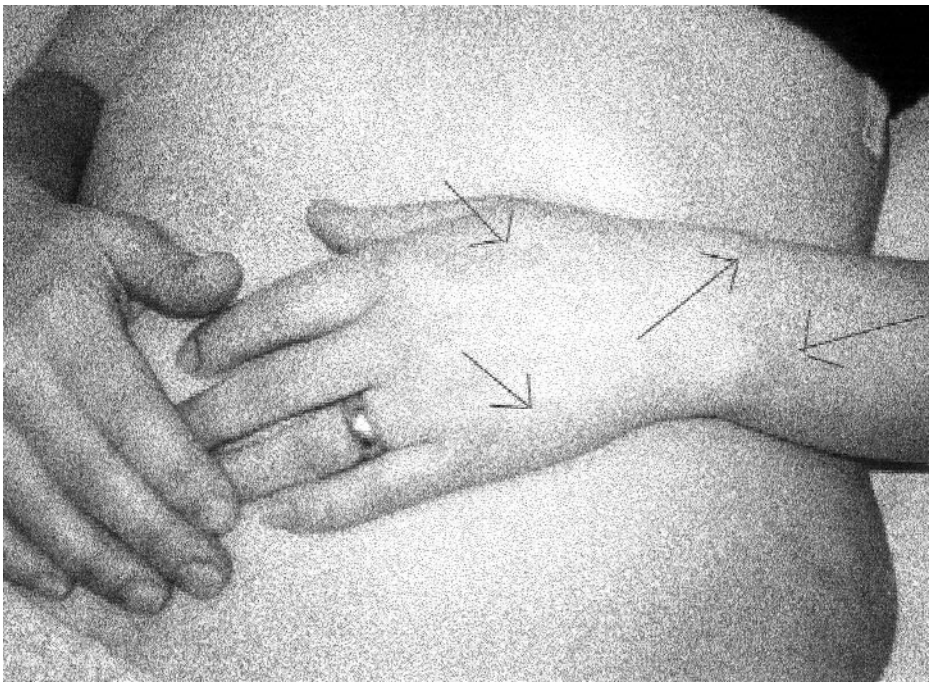

Figure 2. Case 1. Patient had no perception of pain and no withdrawal response, despite the deep imprints of the algometer tip. Arrows: marks after testing of algometric pressure thresholds in the anesthetic limb.

picture of the NDSD characteristics in terms of onset, evolution, and spatial and temporal properties. (b) Sensory examination was performed by one examiner. While the question of test-retest reliability arises, variability is inherent to some NDSD, as described. In 2 other studies[5,6], 2 independent examiners confirmed the presence of NDSD in much higher prevalence rates than reported here.

The NDSD and non-NDSD subgroups seemed to differ in some aspects, but were also similar in a number of variables. However, differences in sex and ethnic background raise the question of sex and cultural variables contributing to the occurrence of NDSD. The highest likelihood of work injury in the NDSD subgroup may be a consequence of higher numbers of non-Canadian males involved in laboring jobs.

There is only limited empirical research documenting the presence/prevalence of NDSD in chronic pain populations. In patients with diffuse myofascial pain, Fishbain, *et al*[5] reported on "non dermatomal sensory abnormalities" (equivalent to our definition of NDSD) in a sample of 247 consecutive patients, seen in one year at a comprehensive chronic pain center, diagnosed with myofascial pain syndrome. NDSD were observed in 40.4% of all patients. Age, workers' compensation issues, and psychiatric diagnoses accounted for only 22.1% of the variance. The authors suggested that physiological factors may have contributed to these deficits and recommended that future studies point to relationships between physiological phenomena and psychiatric pathology. Recently, Kaziyama, *et al*[6] found that 38.2% of 76 patients meeting the American College of Rheumatology (ACR 1990) criteria for FM (chronic widespread pain) presented with hemibody hypoalgesia to pinprick at the side of more intense pain complaints. In patients with neuropathic pain, ipsilateral hemisensory or quadrotomal NDSD were documented in 50% of 24 patients with CRPS (complex regional pain syndrome, formerly known as RSD)[8]. The authors attributed the phenomena to functional alterations in central processing of noxious events.

Regarding the nature of NDSD, hemianesthesia has long been associated with hysteria. "Hysteria" was derived from the Greek word for "uterus" by Hippocrates to denote a female condition caused by "movement of the uterus in the body." Hysterics were treated as witches during the Middle Ages; many were burnt or hanged[9]. Symptoms include "paralyses, muscle contractures, anesthesias and diminished sensibilities, ... partial or total blindness, mutism, various pains, nausea and vomiting, anorexia and spontaneous hemorrhages or, conversely, the absence of bleeding when cut"[10]. After the 17th century the focus changed from the uterus to the brain[11]. Our understanding of hysteria advanced particularly in the late 19th century with Charcot and his pupils (Richer, Janet, Babinsky), while Freud was also strongly influenced by Charcot[12]. Charcot recognized that hysteria occurred in both men and women and appreciated the interplay of psychology in all manifestations of nervous illness. He considered hysterical hemiplegia, paraplegia, or hemianesthesia excellent examples of physiolog-

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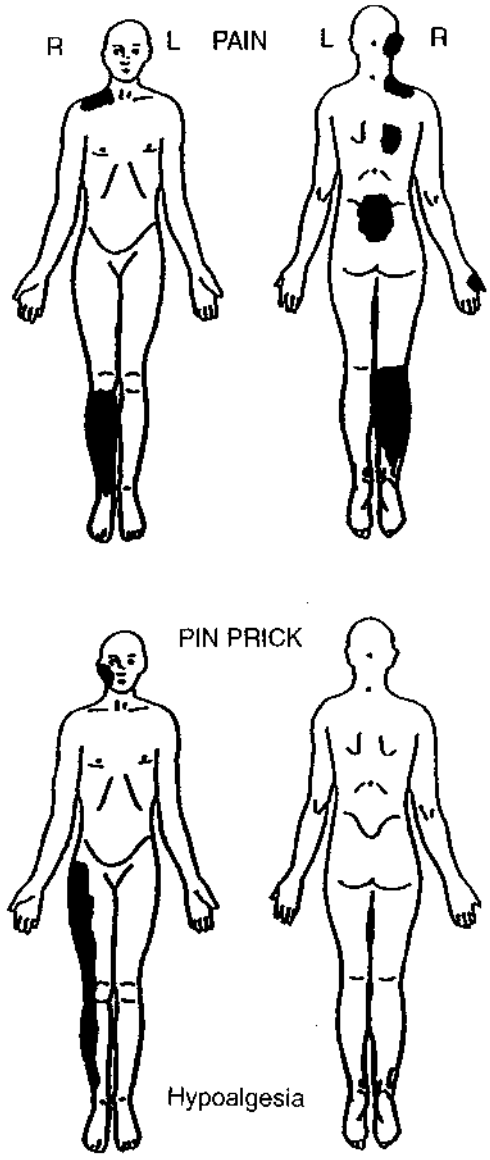

Figure 3. Case 2. First visit, 2.5 years after a minor car accident. Top: patient-drawn pain map. Bottom: deficits to pinprick detected on sensory examination.

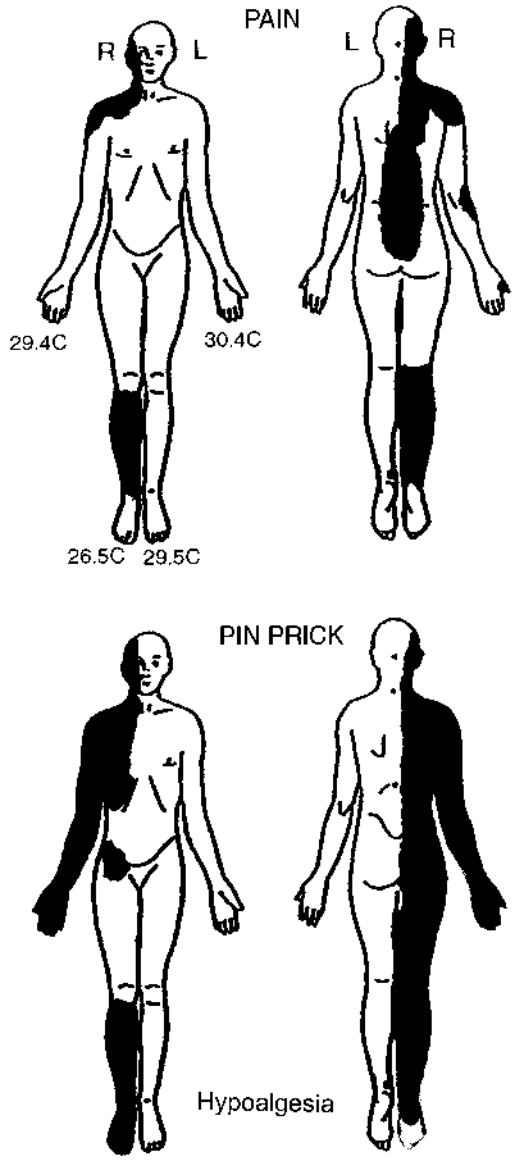

Figure 4. Case 2. Second visit, 2 years after visit 1. Top: patient-drawn pain map. Bottom: marked hemibody hypoesthesia to pinprick detected on sensory examination.

ical dysfunction of the same brain regions that are affected in cases with static anatomical lesions, and he interpreted them as clinical expressions of "dynamic" or physiological aberrations in neural function[10].

Moldofsky and England[13] studied 5 hysterical male subjects with minor but intensely frightening industrial accidents, presenting with hemianesthesia or hemihypoesthesia, motor weakness, and pain (4/5 patients). Scalp somatosensory average evoked responses were measured with strong tactile stimuli, and instead of habituation contralateral to the affected side, facilitation was found. The authors proposed involvement of a spinal gating system, associated with the "unusual" cognitive state of these patients.

The dynamic nature of NDSD was first observed by Moriwaki, *et al*[14], who reported progressive "shrinking" of hypoesthesia surrounding an area of allodynia in tandem with pain relief after variable therapeutic interventions in a population of neuropathic pain patients. In 1997, our group reported the responses of 11 patients with lateralizing pronounced somatosensory deficits to intravenous infusion of placebo (normal saline) controlled SA[3]. This medium action barbiturate has served as an important diagnostic tool in our unit for 15 years[15,16]. The patients had either definable pathology inadequately explaining their complaints or absence of any detectable peripheral pathology after multiple investigations. Complete and permanent (at 2 yrs

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followup) resolution of longstanding hemisensory deficit and pain occurred with normal saline infusion in one patient. Both NDSD and pain improved dramatically with SA but not normal saline infusion (lasting hours) in 6 other patients, while pain improved but sensory deficits persisted in the remaining 3 patients, and both pain and sensory deficits were unchanged in one. Personality profiles obtained in 8 of these patients showed deep MMPI-II "Conversion V" profiles and specific MCMI-II (Millon Clinical Multiaxial Inventory) scale elevations. We proposed that a psychobiological substrate at cortical or subcortical levels was responsible for these deficits, possibly associated with a certain type of personality organization. Recently, we also described dramatic but shortlived reversal or elimination of dense and diffuse sensory deficits during SA infusion in 2 additional patients with extensive syringomyelia[4]. In these cases we observed that dynamic deficits can be superimposed on structural neurological abnormalities.

Possible Neurophysiological Correlates of NDSD and Directions for Future Research

In terms of levels of central nervous system involvement, despite their dynamic nature, the resemblance of NDSD with structural sensory deficits is striking. Hemisensory deficits have been described after large lesions of the contralateral parietal cortex, lesions of the thalamus, and other levels of the neuraxis; quadrotomal or whole limb deficits occur also in complete brachial plexus deafferentation. Apart from the sensory abnormalities, in structural lesions such as brachial plexus avulsion or central post-stroke pain syndrome, pain persists often in the presence of complete deafferentation. In subjects in this study, pain has persisted despite suppression of cutaneous and very often deep sensation. We speculate, therefore, that NDSD constitute an unsuccessful and "maladaptive" attempt of the central nervous system to "shut down all peripheral inputs" in an effort to "shut down" or control pain. Thus, NDSD seem to be examples of "functional deafferentation" resulting from maladaptive neuroplasticity.

Both pain and NDSD are modifiable under intravenous SA infusion[3]. Our current knowledge of the pharmacology and effects of SA on pain and sensory abnormalities has been summarized[7,16,17]. However, it is unknown what pharmacological actions of SA are involved in the normalization of NDSD. Detailed speculation about possible neurobiologic circuitries underlying NDSD and other pain phenomena is beyond the scope of this paper. Our preliminary functional MRI data from patients similar to those reported here[18] indicate that these patients do not activate their somatosensory cortex after non-noxious or noxious stimulation of the symptomatic limb.

Clinicians who see patients with NDSD should be mindful of their association with psychological factors. Persistent pain and disability may be attributed to the presence of undisclosed/hidden pathology and/or the possibility of "sensitization" of the central nervous system after the original injury; the latter concept is based primarily on experimental animal work[19]. Pain and disability may be associated with psychological/personality factors, job dissatisfaction, the presence of compensable issues, etc[20]. It is conceivable that dynamic aberrations of brain functions can occur under a multiplicity of emotionally charged conditions and/or in certain personality organizations where the individual utilizes specific mechanisms to avoid unpleasant physical and/or emotional events. The magnitude of the original physical trauma and duration of actual nociception may be insignificant, but serve as a trigger of underlying central mechanisms in emotionally charged personal or psychosocial situations. This study and others[21] in patients with NDSD and diffuse pain in the absence of detectable peripheral pathology support the popular view that NDSD patients demonstrate more intense and abnormal pain behavior compared to non-NDSD subjects. However, it is unclear from our study what biological or psychological factors discriminate NDSD patients from non-NDSD patients, given their considerable similarities. This question should constitute the focus of future research.

In a recent paper reporting 14–22% prevalence of chronic widespread pain in the general population by American College of Rheumatology criteria for FM, the overall prevalence of mental disorder in the chronic widespread pain group was 16.9% versus 6.5% in those without diffuse pain[22]. One wonders what could be the prevalence of NDSD in the general population with and without diffuse pain and/or mental disorder.

It should be stressed that these deficits are not limited to subjects with compensable issues or patients with minimal or no detectable peripheral pathology (Mailis, *et al*, unpublished observations). We have seen NDSD developing over time in numerous cases of localized nociceptive or neuropathic pathology, at which point pain may spread and become unresponsive to traditional treatments. Therefore, it would be inappropriate to conclude from our observations (our study targeted a specific group) that NDSD develop only in the absence of peripheral detectable pathology. Systematic studies of psychosocial factors of NDSD patients are currently under way.

Our study reveals a high prevalence of NDSD in a specific chronic pain population. It also suggests that intractable pain in the presence of no detectable or minimal peripheral pathology may be associated with a central neurobiological substrate and psychological/personality factors. Our experience has convinced us that the development of NDSD is a bad prognostic sign for response to treatments. In terms of treatment implications, patients with diffuse pain complaints often undergo multiple treatments (multidrug therapy, with occasional narcotic analgesics, variable physical modalities, trigger point, nerve and periph-

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eral joint blocks, etc.) or to the contrary, the patients' complaints are dismissed as mental/psychological disturbance or malingering. Our study provides an intellectual framework, based on the biopsychosocial concept of illness and pain rather than the dualistic "medical" model of disease, by suggesting an interplay of psychological/personality factors and original physical trauma (often insignificant), which in turn mobilizes/generates central abnormalities expressed in sensory (and very often motor) deficits.

Further research is necessary (a) to elucidate the prevalence of NDSD in the general population with chronic widespread pain versus specialized pain clinic populations, with or without litigation or other psychosocial issues; (b) to document the evolution of these deficits before and after resolution of compensable issues or psychological stressors; (c) to investigate their neurobiological substrates with functional neuroimaging and other techniques; (d) to shed light into personality predisposition and/or psychological factors associated with NDSD; and (e) to address treatment approaches.

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