

Chronic regional pain syndrome without demonstrable pathology induced by minor trauma: Formulation of an explanatory diagnosis

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Abstract

Diagnoses frequently fail to meet standards which enable understanding of the case sufficient for statistical prevalence requirements, optimal management, prognostication, and communication with the patient. This is particularly evident in medico-legal contexts, as illustrated by the case history of a claimant with chronic pain-related disability but no observable pathology following a relatively minor motor vehicle incident. In cases where pain is the dominant symptom, additional to application of published authoritative principles of diagnosis, the evolving diagnostic methods of pain medicine provide helpful guidance. These include the biopsychosocial conceptual framework, evidence-based stimulus-response somatosensory testing for neuropathic and nociplastic pain phenotypes, and the new pain terminology for the classification of chronic pain developed by the International Association for the Study of Pain Task Force for the International Classification of Diseases-11. This case is presented to illustrate current diagnostic formulation by application of these pain medicine methods. By identification in medico-legal cases of biopsychosocial risk factors for adverse pain and adverse psychological outcomes which are related and unrelated to the injury, the common problem of bias is reduced.

Keywords

Chronic pain classification; Diagnosis Medico-legal; Biopsychosocial evaluation; Somatosensory testing.

Abbreviations

CT scan: Computed tomography; ICD: International classification of diseases; IEQ: Injustice experience questionnaire; MRI: Magnetic resonance imaging; MVA: Motor vehicle accidents; ODG: Official disability guidelines; PCL-5: Posttraumatic stress disorder checklist for DSM-5 (Diagnostic and Statistical Manual of Mental Disorders).

Introduction

This case illustrates the challenge of a meaningful diagnosis in the common circumstance of an insurance claimant who reported chronic regional pain with disability after relatively minor injury and had no imaging-revealed pathology. The medico-legal consultant, reporting to the insurers 10 months after the injury, made an initial diagnostic statement that she may have sustained a whiplash type injury to her neck and back following an incident in a motor vehicle. He did not consider that she had sustained permanent injury and could not rule out psychogenic potentiation or non-organic factors (not defined). Her whiplash (injury) “should have resolved as per ODG Guidelines (Official Disability Guidelines published by the Work Loss Data Institute, used by the majority of Australia’s insurance providers. Relevant outcome measures include percentage of care resolved, evidently interpreted as injury resolved)”. The final diagnosis was age-related degenerative changes in her cervical, thoracolumbar spine, and it was acknowledged that, unfortunately, there were no imaging reports available to confirm this diagnosis. Subsequent imaging revealed no significant degenerative or post-traumatic spinal changes.

The limited, essentially biomedical, initial diagnostic statement (whiplash injury) referred to the mechanism of injury. The final diagnosis, influenced by an assumption that an injury of this type should have resolved, attributed the ongoing pain and related disability speculatively and erroneously to aggravation of degenerative changes. The diagnosis was not made in accord with current best practice [1-3]. The diagnostic assessments did not validly explain the reason for ongoing pain-related disability and were thus an inadequate basis for therapeutic guidance and prognostication.

The cited references to best practice for medical diagnosis [1-3] are authoritative, and medical practitioners should aspire to the principles and diagnostic process. Although patient-centered approaches and teamwork in diagnosis, including the patient and family, were encouraged in these publications, the authors did not specifically include the biopsychosocial conceptual framework in formulating a diagnosis and assessment, notwithstanding recommendations to do so in medical practice [4,5], pain medicine [6-10], occupational and medico-legal practice [11-13]. It is acknowledged that the implementation of biopsychosocial assessment for management has not consistently met expectations [10], commonly remaining largely biomedical, but that does not invalidate the principles.

The advantages of pain medicine principles and practice for informative diagnosis in cases with dominant pain and related disability include:

- The overall patient-centered biopsychosocial conceptual framework (Sir William Osler: “...the good physician treats the disease, the great physician treats the patient who has the disease”)
- The multiaxial formulation of the diagnosis informs on pre-and post-injury/illness medical risk markers, sleep impairment, psychological and social factors which influence the symptoms and disability. Such diagnostic formulation is essential to optimal management and prognostic assessment. Valid judgement essentially depends on knowledge and on minimising bias [14], and these principles, critical in medical and in medico-legal practice, are potentially achievable through the pain medicine diagnostic process [15].

- Stimulus-response somatosensory testing, as applied by the author in research and clinical practice [16-19, Protocol in Supplementary information] is informative for pain phenotyping, implications of central sensitization, and mechanistic phenotyping (nociceptive, neuropathic and nociplastic pain) [20], especially in chronic pain contexts.

Evidence in support of the practicality of these concepts and processes is available [21] and has been contributed to also by a recent publication and citations therein entitled “Biopsychosocial outcomes of chronically painful injuries sustained in motor vehicle accidents: a survey of 300 claimants” [19]. The conclusions were: “Biomedical factors, including older age, impaired sleep and indicators of widespread central sensitisation, and psychological factors, including stress, anxiety and depression, were the most prominent multivariate associations as statistical predictors of major adverse sequelae (including permanent unfitness for work and serious social decline) contributing to non-recovery for claimants with chronic pain post-MVA. The multiplicity, complexity and interacting nature of the sequelae to even mild or moderate injuries sustained in motor vehicle accidents require early and comprehensive biopsychosocial assessments for optimal management.”

In this case report, the focus is on diagnosis rather than assessment. The former is the identification and naming of a disease, illness or condition, determined by a medical practitioner. There is much more to diagnosis, as outlined in the Discussion. The latter is an evaluation by a healthcare practitioner of symptom severity and disability to enable an informed decision about such matters as fitness for work, provision of care and assistance, compensation, and what therapeutic initiatives to apply. Diagnosis and assessment tend to overlap or to be combined [22].

Case Presentation

A 51-year old woman, a registered nurse, presented in May 2022 for medico-legal assessment and report, claiming injuries sustained in an incident involving a motor vehicle two and a half years earlier. The history is abbreviated.

Pre-injury factors potentially predisposing to adverse injury-related pain and psychological Outcomes: The pre-accident factors that probably conferred vulnerability to adverse pain and adverse psychological experiences from injury (risk markers and risk factors) included family history (father was stated to have had bipolar mood disorder and sister had fibromyalgia syndrome), sexual abuse as a child, a history of anxiety and depression, work related stresses, iron deficiency, vitamin D deficiency and the painful phenotype of restless legs syndrome. The biomedical predispositions to adverse response to injury included a history of relatively minor recurrent cervical spinal pain with inconsequential observations on radiographs. There was a more prominent history of recurrent lumbosacral spinal pain dating from a minor injury in 2017, with CT scan evidence of degenerative changes in the form of osteophytes and facet joint arthritis especially on the left involving the L5/S1 facet joint.

The recurrent low-grade pain in the lumbosacral spine as with the cervical spine was not primarily caused by these degenerative changes but rather biomechanical factors impacting, in the lumbar spine, on

the low-grade degeneration.

There had been significant work contexts that contributed importantly to the ongoing mood disorder and anxiety. She referred particularly to workplace bullying. Leading up to the time of the accident she was working as a registered nurse at a community health centre which was a happier environment.

The injury and sequelae: In October 2019, the claimant was a front seat passenger with seatbelt applied in a car being driven by a nurse colleague. The driver required urgently to do a U-turn, an abrupt decision while travelling at 80 km/h. The car crossed the median strip, which was a bumpy ride, and then entered the road travelling in the opposite direction. The bottom of the vehicle, seemingly directly under her, struck a cement portion of the median strip causing her to rise up and her body jerked to the left striking the door with her left shoulder. She remembered bracing but did not recall immediate pain. She was frightened by the whole experience but did not remember specific fear of injury.

That night at about 3.00 am, she wakened feeling a lot of pain in the left side of her neck posterolaterally, extending towards the left shoulder. Such was the intensity, she felt nauseated and vomited. It was not until two days after the incident that she was able to consult her general practitioner. The initial diagnosis was “whiplash injury to neck and upper back” with spasm in neck muscles radiating down to left upper back/scapular region and decreased range of motion of her neck/cervical spine, about half normal range in all directions, very little forward flexion. There were indications of neuropathic symptoms, interpreted as radiculopathy, extending down her left arm to her hand in C7/8 dermatomal distribution. The initial management plan was analgesia.

Left thoracic and cervical spinal pain with headaches, lumbar spine pain with leg pain to her foot persisted. Intercurrent problems which might have aggravated her symptoms included: anxiety with panic attacks requiring sertraline, low serum iron biomarkers and low vitamin D. She is a vegan and had over a long period taken supplements of iron, vitamin D, vitamin B12 but was only taking iron supplements in a multivitamin product. During the past 2 years she had symptoms consistent with painful restless legs syndrome. She was highly anxious in traffic and would only travel in a car driven by her partner. She experienced flashbacks or vivid re-experiences of jolting, bad dreams about sudden stopping and being thrown, and she was also hypervigilant, but was not assessed for post-traumatic stress disorder.

During 2020 she underwent treatment by a pain medicine specialist and a sports medicine specialist, additional to chiropractic and physiotherapy. The lumbosacral spinal and referred pain had settled to pre-MVA levels of discomfort. There were issues with bullying from managers, and she was stressed by a family concern. By June 2020, she was managing only three hours work on two days per week. Impaired sleep was a major factor in her limited work capability including lack of confidence in nursing decisions. She was not capable of managing her domestic requirements and had two friends who assisted. She maintained sertraline and minor analgesia. Symptoms remained stable, but more favorable work enabled increase in hours.

Psychological testing: In June 2022, her self-report scores on the PCL5 Questionnaire for post-

traumatic stress disorder, Pain Catastrophising Scale, Depression, Anxiety and Stress 21 (DASS 21) scale, and IEQ questionnaire for perception of injustice were surprisingly low. After discussion, she acknowledged deliberately reporting low to reduce any perception that, perhaps as a nurse, she might be considered to have psychologically dominated symptoms. Her summed score on the Örebro Musculoskeletal Pain Screening Questionnaire (Short), probably reliably reported, was a modest 47/80. Impaired sleep, with symptoms of restless legs syndrome, was importantly limiting her capacity to work.

Imaging investigations: In December 2019, MRI cervical spine showed only that at C4/5, C5/6 and C6/7 mobile segments there were very minor (inconsequential) disc bulges. The cervical spinal cord showed normal signal intensity. There was no evidence of significant facet arthropathy in the cervical spine. In the thoracic spine, the intervertebral discs were of normal height and signal intensity without disc bulges or herniation. There was no evidence of degenerative change or traumatic pathology. In the report of the radionuclide bone scan of February 2020, there were no signs of significant degenerative or post-traumatic abnormalities. The radionuclide bone scan of February 2020 showed no signs of significant degenerative or post-traumatic spinal pathology.

Examination: Mainly positive observations are recorded here. The patient/claimant appeared very tired and agreed with that assessment, consistent with impaired sleep as a major issue. She reported feeling predominantly left sided pain from the suprascapular region, extending down her posterolateral thorax. There was some pain at the base of her neck posterolaterally. She said that her left arm did not feel quite normal, but it was difficult to describe. The worst site of pain was in the region of her left scapula. Although overweight, she said that she had lost 15kg since the motor vehicle incident of October 2019.

There was a full range of cervical spinal motion but with some end-range pain in full left rotation felt in the lower left lateral paraspinal region (a little discomfort on the right side on full right rotation). Left lateral flexion brought on left posterolateral thoracic pain.

While she was lying prone, stimulus-response somatosensory testing [Supplementary information] was performed. Responses to light touch (brush) stimuli, both static and especially dynamic, were abnormal extensively on the left side of her body. The hyperaesthetic responses were elicited mildly throughout her left upper limb and throughout the left thoracic back to the upper lumbar back region. Responses to punctate pressure stimuli (Neurotip or Neuropen) were abnormal throughout her left hemibody including face, neck upper and lower limb extensively, back from suprascapular region to buttock, and abdomen. Within that distribution, especially left thoracic back, there was facilitated temporal summation in response to repetitive punctate pressure stimuli. The right side of her neck, posterolaterally, was also hypersensitive. She volunteered that some of the sites, notably the left posterolateral thorax were persistently uncomfortable well after ceasing the stimuli. Responses to cold stimuli (Thermoroll at 22°C) were also extensively hypersensitive/hyperaesthetic on the left side but not in full hemibody distribution.

Responses to deep pressure stimuli were abnormal in very wide distribution, virtually throughout her body with only slight tendency in some regions to be more abnormal on the left side than the right side. There was widespread low pressure pain threshold and widespread elicitation of temporal summation in

response to repetitive deep pressure at the pain threshold. *Comment:* These left hemibody cutaneous sensory abnormalities and widespread low pressure pain threshold and facilitated temporal summation were indicative of disordered sensory processing in the central nervous system with features implying central sensitization, extending well beyond the pain region.

Finally, there was low pressure pain threshold (tenderness to deep palpation) throughout the cervical, thoracic, and lumbar vertebrae. At the end of the examination, she volunteered that she had a headache which was quite extensive and symmetrical and notably included the right and left side of her forehead.

Discussion

Best practice clinical diagnosis: In the beginning, we should heed the guidance of Sir William Osler: «Listen to your patient – he is telling you the diagnosis.» and «Medicine is a science of uncertainty and an art of probability.» Not infrequently, current consultations begin with the patient presenting a diagnosis from internet and other sources. While the patient is usually endeavouring to tell the perceived truth, a quote from Douglas Murray [23] is apposite, perhaps particularly in medico-legal contexts, “Lived personal experience can never be fully comprehended, but neither can it wholly be believed.”

There are various categories of diagnosis, including provisional, pathological, laboratory, radiological, and differential diagnosis, but the focus of this case report is on the best achievable clinical diagnosis after review of all the facts. Sound judgement is critical, and that requires knowledge and the minimising of bias [14]. The diagnostic process begins with gathering data, integrating it, and formulating a diagnostic hypothesis, ideally leading to iterative Bayesian thinking [3]. A diagnosis based on patient centeredness [2] and multiaxial and biopsychosocial conception [15], which commonly incorporates pattern recognition, is supported by teamwork and by pathological and/or imaging investigations, and which addresses causal influences, provides the optimal explanation for the patient’s symptoms and disability and thus is a sound guide to management, prognostication, and communication with the patient. A diagnosis is rarely final and usually needs to be tested and verified over time [2]. Sometimes algorithms or clinical decision support systems are applied.

In medical and medicolegal practice, wholly or verifiable objective evidence would be preferred, such as post-traumatic pathology. Unfortunately, the reality is that objective evidence after trauma is not present in many cases and where present might not be reliable (*e.g.* a single level of degenerative change in the spine but symptoms and signs arising from multiple levels). Furthermore, it has been very clear in multiple studies that there is often little correlation between pathology and chronic pain-related disability. Chronic pain diagnosis according to the International Classification of Diseases-11 (ICD-11) [24] and the neurobiological pain mechanistic categorization [20], which is integrated with the psychological and social factors, better account for the patient’s history of pain and for the evident related disability.

It might be suggested that pain medicine has a large subjective or intersubjective (shared experience between patient and doctor) component. However, evidence-based pain medicine is at a higher level, frequently applying published methods which are widely accepted by consensus, *e.g.* of pain me-

dicine scientists and clinicians. In this case, as discussed below, the clinical criteria for nociplastic pain augmenting the post-traumatic pain disorder were met. Such consensus evidence has utility, for example for understanding, management and communication. Apparent objective evidence, such as post-traumatic intervertebral disc lesion on imaging investigations, provides qualified truth regarding pathology but has limited utility for management since, for example, the pain may be arising in a region or multiple sites and be augmented by central sensitisation and diminution of descending pain modulation systems as well as by impaired sleep and psychological factors.

The reality is that the aspirational standards of diagnosis are rarely achieved and were certainly not achieved in the medico-legal report to the insurer as summarised in the introduction. The author's medico-legal diagnosis also did not attain the highest standard, hence this case report is an attempt to meet and illustrate as many of the optimal requirements as feasible. In the medico-legal context, there must be strong emphasis on causal interpretations and on the necessity to minimise bias.

Clinical diagnosis in this case: The diagnostic formulation conventionally begins with biomedical determination. There was no observable injury-related pathology. The International Association for the Study of Pain Task Force for ICD chronic pain classification decided to give first priority to pain etiology, followed by underlying pathophysiological mechanisms (clinical phenotyping), and finally the body site. The pain disorder was caused by the trauma experience within the motor vehicle in October 2019, did not resolve but rather extended in anatomical distribution, remained the dominant symptom, and although contributory factors were identified, no alternative causal evidence occurred. **The biomedical musculoskeletal/spinal predispositions to adverse response to injury** included a history of relatively minor recurrent cervical spinal pain with inconsequential observations on radiographs. There was a more prominent history of recurrent lumbosacral spinal pain but that did not prove to be highly relevant after two years.

This pain disorder is categorised in ICD-11 as posttraumatic pain [24,25]. Most pain associated with whiplash injury, the most relevant subcategory of posttraumatic pain discussed by Schug et al [25], does not fulfil the current diagnostic criteria of neuropathic pain (being primarily nociceptive), but central hyperexcitability plays a major role in the pathogenesis. The stimulus-response somatosensory test responses in this case were consistent with extensive central hyperexcitability (including central sensitisation), referring especially to hypersensitive responses to punctate pressure and to deep pressure stimuli, with temporal summation of pain responses to repetitive stimuli, along with persistent after-sensations.

Features of secondary or derivative central pain mechanisms were identified as nociplastic [26,27] and contributed to the chronicity, extended anatomical distribution and several associated symptoms. Applying the International Association of Pain criteria for nociplastic pain [26], while acknowledging that they are probably not yet settled [27,28]:

1. The pain was chronic, regional in distribution, with no confirmed evidence of peripheral neuropathic pain.
2. There was a history of pain hypersensitivity in the region of the pain (and extending beyond), and she

had sensitivity to touch, to pressure, to movement and to cold.

3. There were comorbidities (any one of the following is required) which included sleep disturbance, fatigue, cognitive problems (memory and focus attention) and increased sensitivity to sound as well as to some extent to smell and to light.
4. Evoked hypersensitivity was elicited from regions of pain (and extending beyond), although she did not have static or dynamic mechanical allodynia nor heat or cold allodynia. Rather, she had responses indicating cutaneous hyperalgesia to punctate pressure and to deep pain stimuli, implying central sensitization. She had painful after-sensations following such stimuli.

Personal and environmental (especially psychosocial) factors contributed importantly to the pain experience and related disability, and while some factors may be classifiable as assessment, they contribute critically to the diagnostic formulation, including the pain mechanisms, especially relevant for the medico-legal diagnosis. Furthermore, identification of such factors which are unrelated to the injury demonstrates consciousness of minimizing pro-claimant bias in the medico-legal context.

First and foremost, the patient/claimant had seriously impaired sleep which is a known risk factor for augmented pain and related disability and for emotional problems, and which interfered with her capacity for work. Impaired sleep was partly the consequence of pain but was also prominent pre-injury. She had a history of iron deficiency which predisposes to impaired sleep [29] and to pain sensitivity and vulnerability [30]. Restless legs syndrome was symptomatic pre-injury, adversely influenced sleep, was probably causally influenced by iron deficiency [31] and has associations with multiple pain disorders (probably involving shared causal factors) [32,33].

Other pre-injury factors that probably conferred vulnerability to adverse pain and adverse psychological experiences from injury (risk markers and risk factors) included family history (father was stated to have had bipolar mood disorder and sister had fibromyalgia syndrome), sexual abuse as a child, a history of anxiety and depression, work related stresses, and vitamin D deficiency.

Post-injury psychosocial factors which potentially aggravated pain, sleep and mental health, most consequential on the motor vehicle incident and others unrelated, included: early post-traumatic stress, work related stresses, family concerns, medicolegal issues and a strong perception of unhelpful behaviour by the insurer's representatives, loss of capacity to work fulltime and thus financial stresses.

Conclusion

The patient/claimant developed a chronic regional (left side of neck, left shoulder and posterior hemithorax) pain disorder after minor, mainly indirect, injury in a motor vehicle incident. There was no observable pathology. Two and a half years later, the medico-legal challenge was to provide a diagnosis which best explained the ongoing pain, related disability, reduced capacity for work, psychological symptoms and social outcomes. Although there would inevitably be differing beliefs and contentions, it is suggested that the principles and diagnostic process of pain medicine enables diagnostic categorisation, and provides an

appropriate basis for management, prognostication and communication with the injured person.

After diagnostic formulation, in summary, this injured woman had a posttraumatic chronic regional pain syndrome with nociceptive and nociplastic phenotypic pain features. The disability and impact on life roles, including limitations for work and social function, had been extensively contributed to by pre-injury and post-injury biomedical, psychological and social factors. The injury, although assessed as relatively mild, impacted on a vulnerable individual and had a substantial causal influence on chronic pain and on her multiple adverse outcomes.

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