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Research Article

Sudden-Onset Widespread Body Pain (Fibromyalgia) with or without an Inciting Event: A Case Series

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Abstract

Background: The presence of sudden-onset fibromyalgia (FM) is poorly understood, characterized, and appreciated in both previous literature and the clinical setting. In this case series, we present 10 cases of sudden-onset FM seen in a community-based pain clinic, to characterize the presentation of this condition, stemming from both external trauma and idiopathic etiology.
Methods: This retrospective case series identified 10 patients diagnosed with FM attending the pain clinic. These patients were referred to chronic pain management clinic in Thunder Bay, Ontario, Canada, from January 2019 until December 2021 and met the diagnostic criteria for FM. Information was collected on sex, gender, age, details of signs and symptoms, and FM severity score as well as clinical findings and outcomes.
Results: The case series identified 10 community residents (9 women and 1 man, F/M: 9/1, age range: 34-64 years, mean age: 51.7 years), with symptoms attributed to FM. Majority of patients suffered from total body pain and mental disorders such as depression. 60% of patients were on opioids at the time of referral. Combination of pharmacological and non-pharmacological management improved their pain symptoms within 3-6 months on follow-up.
Conclusion: Overall, effectively identifying sudden-onset FM can help clinicians improve patient-oriented outcomes and avoid the use of unnecessary narcotics in addition to better treating their patients.

Keywords: Case Series; Chronic Pain; Widespread; Fibromyalgia; Sudden-Onset

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Background

Fibromyalgia (FM), also known as chronic whole-body pain with central amplification, is a disorder of the neuromuscular system causing widespread body pain, tenderness, and fatigue. In addition, it commonly affects patients' memory, sleep, and mood (1).

Cases of FM are reported around the world throughout ethnic groups and cultures. In the United States (US) and Canada, FM is most common amongst women over 40 years old and ties closely to low socioeconomic status (SES) indicators and patients who exhibit risk factors for chronic disease (2-4). Most recent studies estimate the overall prevalence of FM at 1.5% in the adult Canadian population (2).

FM has traditionally been characterized as a gradual-onset disease. The presence of sudden-onset FM is poorly understood, characterized, and appreciated in both the literature and clinical setting. However, sudden-onset FM is more common than traditionally acknowledged in clinical practice, with prevalence rates of up to 25% amongst patients suffering from FM (5). In the vast majority of cases, external shock such as trauma or infection is the root insult leading to the development of sudden-onset FM, with little previous research finding idiopathic sudden-onset FM (5). Patients with sudden-onset FM usually undergo numerous investigations and continue to have chronic pain for years.

This case series aims to present a review of 10 cases with sudden-onset FM, with the aim of describing the characteristics and common factors of patients diagnosed with this condition referred to a community pain management program and diagnosed by a pain specialist.

Methods

Setting/Population: This is a retrospective descriptive case series drawn from all new patients referred during a 3-year period to a community-based pain clinic for a diagnostic assessment of their chronic pain (January 2019 until December 2021) (6). All patients included in the study provided informed consent. This study has Ethics Approval from St. Joseph's Care Group, Ontario, Canada (SJCG REB # 2020005).

Inclusion Criteria: We included patients who presented with sudden-onset whole-body pain, had not been previously diagnosed conclusively, had pain in at least four regions out of five (left upper, right upper, left lower, right lower, axial), and were subsequently diagnosed and treated for FM at our clinic from January 2019 until December 2021. Only patients who scored above 13 in the FM severity score [Widespread Pain Index (WPI)≥7 and Symptom Severity Scale (SSS) score ≥5, or WPI 4-6 and SSS score ≥9] and met the diagnostic criteria for FM as per the 2016 revisions to the 2010/2011 Fibromyalgia Diagnostic Criteria by the American College of Rheumatology were included (7).

Data Collection: All patients treated in the clinic had their sex, gender, and age collected routinely at the time of the initial consultation. All patients completed the Brief Pain Inventory (BPI), Patient Health Questionnaire-9 (PHQ-9), General Anxiety Disorder-7 (GAD-7) questionnaires, and a body map of pain areas on intake. Besides, standard

questions were administered to each patient on intake, including inciting events (if any) and associated symptoms (Table 1). Further information was taken from clinical charts for the case series about specifics of FM (FM severity score, WPI, SSS), description of symptoms after the sudden onset of pain, and clinical assessment findings. Additionally, follow-up notes were reviewed to determine patient outcomes (for instance, pain score and functionality) (6).

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Results

The cohort with prolong symptoms consisted of 10 patients (9 women and 1 man, F/M: 9/1) with age range of 34-64 and mean age of 51.7 years. All patients were younger than 65. Sixty percent of patients reported severe pain intensity ranging from Numerical Rating Scale (NRS) of 8 to 10. Of the total cases, 60% of patients were taking opioids prescribed by their family doctors or other physicians. All patients were new referrals to pain clinic and the onset of pain duration ranged from 2 to 31 years when they developed symptoms of FM. The demographic characteristics of the included patients are presented in table 1.

Table 1. Demographic characteristics of included patients

Variables	Value
Average age (year)	51.7
Age range (year)	37-64
Gender (women) (%)	90
Employed (%)	50
Married (%)	70
Average pain duration (year)	14.8
Divorced (%)	20
Widowed (%)	10
Average BPI (pain interference score)	50.9
BPI (pain interference score) range	32-64.70
Associated symptoms	100% of cases complained of chronic fatigue, sleep issues, and poor concentration. 70% of cases had irritable bowel and 50% had urinary frequency.
Most common comorbidities	Depression (60%), migraine (30%), tension headache (50%)
Most common medications at time of visit	50% using prescribed opioid medication, 40% using antidepressant medication

BPI: Brief Pain Inventory

Illustrative Case Reports: While table 2 contains the cumulative results and other information relating to all of the cases, including outcomes after treatment, we provided information on three cases that spanned the whole pathology range (biomedical vs. psychiatric or combination).

Case Example A (Table 2, Case 5)

A 54-year-old woman was referred for widespread body pain. She was well-adjusted, working as an early childhood educator and living at home with her husband and two children. The patient reported that her pain had suddenly begun a year ago and had gradually worsened. She denied any injury, physical, or emotional inciting event. Her pain sites included the lower back, torso, abdomen, and legs, and she complained of regularly interrupted sleep. Her pain interference score on the BPI was 50/70. Her GAD-7 score was 10/21, and her PHQ-9 score was 15/27. Her Index of Education Quality (IEQ) score was 33/48. Before she visited our clinic, she had been sent for multiple investigations, including a full spine magnetic resonance imaging (MRI) which was unremarkable, as well as electromyography (EMG)/nerve conduction studies (NCS) which showed mild nonspecific findings of peripheral sensory neuropathy. She had not been previously given a particular diagnosis for her whole-body pain.

Clinical Impression: This patient's physical examination was not consistent with any radiculopathy or inflammatory radiculopathy, but maybe peripheral sensory neuropathy can explain her feet numbness. However, I cannot explain her hip, lower abdominal, pelvic, shoulder, and lower back pain with the sensory neuropathy. On the FM severity score, she scored 10/19 for WPI and 10/12 for SSS. This was associated with the widespread body tenderness sign and associated symptoms of insomnia, fatigue, irritable bowels, and brain fog convinced me to diagnose her with FM. I organized a referral for total body bone scan and blood work [complete blood count (CBC), thyroid tests, C-reactive protein (CRP), erythrocyte sedimentation rate (ESR), and parathyroid hormone (PTH)] to rule out any underlying pathology which all returned back normal.

Case Example B (Table 2, Case 7)

A 60-year-old woman was initially referred for ongoing lower back pain. She was on Ontario Disability Support Program (ODSP) due to head injury and seizure but had worked in maintenance at a bingo hall and a university 16 years ago. She lived alone, divorced for almost two decades, and had one successful adult child living in Ottawa, Canada. She reported sudden onset of the lower back, arm, and leg pain bilaterally starting four years ago when she woke up one morning, accompanied by ongoing trouble with staying and falling asleep. The patient denied any physical, emotional, or psychological inciting event. Her BPI score was 32/70. GAD-7 score and PHQ-9 score were 6, and the IEQ score was 5. As with our first case, she had been sent for extensive investigations, including MRI and computed tomography (CT) scan, which were unremarkable and showed general nonspecific degenerative changes through the lumbar spine, with no compromise of the exiting or traversing nerve root. She had not been previously given a diagnosis for her whole-body pain.

Clinical Impression: This patient has been suffering from 5 years of chronic lower back and right leg pain without any signs of lumbosacral radiculopathy based on the lumbosacral MRI in 2017 and today's assessment. She has never had EMG/NCS. Since she was diagnosed with appendix cancer, I was wondering about the metastasis and lumbosacral plexus involvement or paraneoplastic syndrome including mono-limb neuropathy. I organized a referral for EMG/NCS and total body bone scan with single-photon emission CT (SPECT) of the lumbar spine which returned back normal. In follow-up session, I noticed multi-site body pain and overall body tenderness. These signs were associated with symptoms of headache, insomnia, fatigue, and poor concentration and the questionnaires convinced me to diagnose her with FM. Given the fact that all symptoms started suddenly, she had sudden-onset FM. I prescribed Cymbalta 30 mg for 1 week then 60 mg once or twice a day.

Case Example C (Table 2, Case 8)

A 59-year-old woman working as a secretary was referred for widespread body pain. She reported sudden onset of whole-body pain, which occurred immediately after her involvement as a passenger in a head-on collision 30 years ago, during which she experienced no head trauma or fracture.

Table 2. Cumulative results and detailed information of included patients

Patient	Whole body pain onset	Sex/age	Occupation	Marital status	Average NRS pain rating	BPI score at intake	Past medical history (list)	Therapeutic intervention after diagnosis of FM	Pain improvement on the first follow-up
1	2014	F/44	Office manager	Married	9/10	59/70	Headaches, amenorrhea, and irregular menstrual cycle	Flexeril 10 mg QHS, increased by 5 mg/day every 7 days up to 10 mg TID, multidisciplinary pain management program	30% (6 months) unresolved, discharged attempting meloxicam, venlafaxine, nortriptyline, pregabalin, gabapentin, desipramine, nabilone, duloxetine, quetiapine, discontinued codeine
2	2015	M/37	Cartographer	Married	5/10	38/70	Left ankle fracture, bilateral knee dislocation, left knee meniscus tear	Nortriptyline 10 mg QHS, increased by 10 mg every 7 days up to 50 mg, multidisciplinary pain management program	100% (2 months)
3	2000	F/57	Jewelry designer at home	Married	8/10	62/70	TIA, migraine, concussion secondary to MVC, cholecystectomy, C-section	Duloxetine 30 mg, increased to 60 mg after 7 days, multidisciplinary pain management program	Over 90% (2 months)
4	1999	F/45	Unemployed (previously secretary)	Divorced (living with a partner)	8/10	56/70	Depression, sinus tachycardia, hypertension, migraine, T2D, granuloma annulare, ocular migraines, costochondritis, hyperparathyroidism secondary to vitamin D deficiency, C-section, laparoscopy, cystoscopy	Pregabalin 25 mg BID, increased by 25 mg every 3 days up to 150 mg BID, multidisciplinary pain management program	30% (6 months)
5	2019	F/54	Early childhood educator	Married	5/10	50/70	Depression, vitiligo, lichen planus, lichen sclerosis	Previously on duloxetine, increased to 120 mg, multidisciplinary pain management program	Over 80% (1 month)
6	2016	F/53	Unemployed (disability)	Married	3/10	40/70	Breast surgery, left knee meniscus repair, C-section twice	Previously on duloxetine, increased to 60 mg for 1 month, then 90 mg the next month, multidisciplinary pain management program	Over 50% (3 months)
7	2015	F/60	Unemployed (disability)	Divorced	5/10	32/70	Cholecystectomy, hysterectomy, asthma, depression with suicidal attempt, ulcerative colitis, osteoarthritis	Duloxetine 30 mg, increased by 30 mg every 7 days until symptoms controlled or 60 mg BID, multidisciplinary pain management program	80%-90% (3 months)
8	1990	F/59	Secretary	Married	8/10	64/70	Depression, hysterectomy, breast reduction and cosmetic surgery, OCD, carpal tunnel surgery	Clomipramine 10 mg with the instruction of increasing by 10 mg up to 50 mg, if needed or tolerated, beginning to wean off oxycocet use, multidisciplinary pain management program	50% after weaning off opioids (4 months)
9	1988	F/64	Unemployed (disability)	Widow	7/10	64/70	Depression, anxiety, chronic fatigue syndrome, osteoarthritis, asthma, recurrent pneumonia, chronic bronchitis, bilateral carpal tunnel surgery	Pregabalin 25 mg BID, increased by 25 mg every three days up to 150 mg in the morning and 225 mg at bedtime, beginning to taper off oxycodone, multidisciplinary pain management program	Over 90% initially resistant to tapering opioids, requiring three visits to begin tapering and switch to non-opioid management
10	2003	F/44	Unemployed (due to pain), previously working as a vice principal of an elementary school	Married	8/10	44/70	MI, migraine, dysmenorrhea, depression, anxiety, obesity, T2D, hypertension, iron deficiency with thalassemia trait, hypothyroidism	Pregabalin 25 mg BID, increased by 25 mg every 3 days up to 150 mg BID if needed or tolerated, multidisciplinary pain management program	Over 30% (10 months)

NRS: Numerical Rating Scale; BPI: Brief Pain Inventory; FM: Fibromyalgia; QHS: Daily at bedtime; TID: Three times a day; BID: Twice a day; MI: Myocardial infarction; T2D: Type-2 diabetes; TIA: Transient ischemic attack; MVC: Motor vehicle collision; OCD: Obsessive-compulsive disorder

Her pain was present throughout her large joints, lower back, chest, and neck. She was a well-adjusted, successful woman living with her husband, a daughter, and two grandchildren. Her BPI score was 64/70. GAD-7 score was 17/21 (severe anxiety), PHQ-9 score was 24/27 (severe depression), and IEQ score was 47/48 (very high levels of perceived injustice). Repeated investigations, including CT scan, MRI, and EMG demonstrated generally nonspecific findings though most recent studies showed posterior disc bulging in C4-C5 with right-sided cord flattening but no cord signal abnormality. She had been previously diagnosed with FM but came to our clinic on strong opioid medication as her main form of symptom control.

Clinical Impression: I did not find any sign of radiculopathy, peripheral neuropathy, or inflammatory arthropathy. She has been suffering from widespread body pain, insomnia, fatigue, memory and concentration issues, irritable bowel and bladder, as well as whole body tenderness from the beginning which convinced me to

diagnose her with sudden-onset FM. Her large areas of non-dermatomal somatosensory deficits (NDSDs) across the right side of her body indicates that her pain is "centralized". NDSD has been shown to be associated with maladaptive neuroplasticity. The phenomenon has been described extensively since 2001 by Dr. Angella Mailis and her team at the university of Toronto, Canada, in numerous publications, including functional MRI (Neurology, 2003) that demonstrated clear alterations in brain activation patterns in subjects with NDSDs. NDSDs often present in individuals experiencing both a physical trauma (which could be minimal), as well as psychological trauma (8, 9).

Since she has had some mental health issues, she was prescribed a tricyclic antidepressant (TCA) to address her chronic widespread body pain (FM), insomnia, anxiety, depression, and obsessive-compulsive disorder (OCD). Her family physician advised her to taper off Percocet by decreasing 1 tablet every 7 days until she was off, or rotate to Morphine 30 mg a day and decrease it by 2.5 mg every 7 days until she was off.

Follow-up and Outcomes: All patients reported improvement in their symptoms once FM diagnosis was made and treatment for this condition was initiated. On follow-up inquiry, the patients' self-reported pain improvement on a subjective pain scale followed a bimodal distribution. 80% of patients reported at least 30% pain improvement, with 50% of patients reporting ≥80% pain improvement. In total, 80% of patients gained marked functional ability compared to their baseline function and quality of life (Table 2).

The most common barrier against increased improvement time after diagnosis was continuance of opioid use. Many patients using opioids medication on a chronic level were significantly reluctant to begin tapering, which resulted in a period of lag time before improvement once referred to the pain management program.

Two patients were resistant to treatment. Analysis of their charts did not demonstrate significant common factors between the two. The first had no inciting event or traumatic history, and was living a well-adjusted life with a spouse and regular work. The second had a 27^{+} year history of physical and emotional trauma and a complicated past medical history significant for multiple biophysical conditions.

Discussion

We did a clinical case series over three years and discovered ten individuals with symptoms following the sudden onset of widespread body pain; all were new referrals to our pain clinic. Given that we see 768-864 new patients on average each year attending our chronic pain management program, these 10 new patients with FM account for 0.4% of our yearly referral burden. The results of this study show that the analysis of several case studies, which was used in diagnostic research, provides unique insights into sudden FM experience.

Due to poor presence in the literature, sudden-onset FM can easily be missed or misdiagnosed. As time proceeds, untreated patients with FM tend to develop worsening symptoms affecting the patient's physical, psychological, and social well-being (1).

This report presented 10 cases of sudden-onset FM that had not been previously diagnosed and appropriately managed. As in previous studies, evaluation of the treatment history in these patients revealed that a significant number of patients were receiving opioid medications- especially those patients who had post-trauma sudden-onset FM (10). However, sudden-onset FM can present idiopathically as well. Regardless of etiology, these patients suffered from chronic pain until being diagnosed and managed for FM. We found that FM could present as a sudden-onset condition. If sudden-onset FM is recognized and appreciated more quickly, effective treatment can be started immediately.

We hope that this series will help practitioners raise their clinical suspicion for FM in patients who fit the FM profile and who present with sudden-onset pain. Primary care physicians (PCPs) should carefully review the patient's symptoms and consider the presence of sudden-onset FM in their differential diagnosis. As per the 2016 revisions to the 2010/2011 Fibromyalgia Diagnostic Criteria, a diagnosis of FM does not exclude the presence of other clinically important illnesses (7). Critically, FM is no longer a diagnosis of exclusion and should be on a PCP's clinical radar much sooner than it is currently. Correctly identifying such patients will improve clinical and patient outcomes, by allowing PCPs to reduce unnecessary use of opioid medication, while managing their patients more successfully.

In addition, these cases demonstrate the requirement for further research in the etiology of FM. While traditionally understood as a gradual-onset disease, up to 25% of patients with FM have a sudden-onset disease (5). Such presentations are varied in nature, as demonstrated by our case series. In addition, amongst the sudden-onset category, 86% of such patients have a physical inciting event; our case series presents 60% of patients with the same (5). These findings suggest that there may be a structural component to FM and it is not necessarily a predominantly psychosocially-entrenched disease as traditionally believed (11). Future studies are needed to find the possible causes and gather more data.

Conclusion

Sudden-onset FM should be considered in the differential diagnosis of patients suffering from whole-body pain, especially in the absence of positive rheumatologic or autoimmune screening tests, regardless of onset duration. Correct identification of sudden-onset FM will increase patient-oriented outcomes, reduce over-prescription of narcotics, and result in better utilization of healthcare resources.

Conflict of Interest

The authors declare no conflict of interest in this study.

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