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CLINICAL CASE REPORT

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Oral Burning With Dysphagia and Weight Loss

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Abstract: Fibromyalgia is a disorder characterized by an abnormal pain regulation. Widespread pain, fatigue, and sleep disturbance are the prevalent symptoms. When unusual symptoms are overbearingly predominant at clinical presentation, the diagnosis becomes challenging.

We report on the case of a patient with fibromyalgia, who presented with dysphagia, odynophagia, and glossodynia as prevalent symptoms. Difficulty in swallowing gradually developed over a month prior hospitalization, and worsened progressively so that nourishment and fluid intake were impeded.

Because anemia with mild iron deficiency was found, esophagogastroduodenoscopy was performed, but no lesions were seen in the upper digestive tract. Levels of zinc and vitamin B12 were normal. Intense pain at pelvis and the inferior limbs, which was at a first glance referred to as osteoarthrosis, associated with oral symptoms and feeling of being in the clouds allowed us to diagnose fibromyalgia. Amitriptyline was used, with relief of symptoms.

Although oropharyngeal symptoms were occasionally reported in fibromyalgia, they are often overlooked. The present case, therefore, testifies the need to consider the diagnosis of fibromyalgia when the patient presents with such symptoms that cannot be readily explained on other grounds.

(*Medicine* 94(31):e1163)

Abbreviations: ACR = American College of Rheumatology, CKD = chronic kidney disease, CSS = central sensitivity syndrome, SS = symptom severity, TnI = troponin I, WPI = Widespread pain index.

INTRODUCTION

Fibromyalgia is one of the most common disorders seen by primary care physicians, with a prevalence of 2% in the US population.[1] The incidence of fibromyalgia rises with aging, with a maximum at the age above 60 years, and is typically 7 times more prevalent in women than in men. After several unsuccessful efforts aimed at proving the existence of muscle and connective inflammation, in the 1980s, the term fibromyalgia definitely replaced the older term 'fibrositis.'[1]

Currently fibromyalgia is seen as one of several disorders referred to as central sensitivity syndrome (CSS), which are all characterized by an abnormal pain regulation (Table 1). The overlapping of $\geq$2 such disorders supports the contention that central sensitization is the unifying mechanism of such array of diseases.

Widespread pain, fatigue, and sleep disturbance are the characterizing core symptoms of fibromyalgia. Other common symptoms include cognitive difficulty, headache, paresthesia, and morning stiffness. When unusual symptoms for fibromyalgia are overbearingly predominant at clinical presentation, the diagnosis becomes challenging.

CASE REPORT

An 80-year-old Caucasian woman presented at the Emergency Department because of diffuse pain at the anterior chest and epigastric region, dysphagia, odynophagia, vomiting, and fever. Her blood pressure on presentation was 145/80 mmHg, with a heart rate of 90 beats/min.

Blood tests showed mild increase of white blood cells (12.42×10^9 cells/L) and C-reactive protein (7.2 mg/L). An EKG documented subtle ST-segment depression in V3 to V6 with normal troponin I (TnI) levels. An abdomen X-ray failed to show signs of bowel obstruction or free intra-abdominal gas. Chest X-ray was unremarkable. Nausea and vomiting disappeared after 10-mg intravenous metoclopramide. Because of her history of coronary artery bypass grafting, and the persistence of pain and dysphagia, she was referred to our internal medicine unit.

Assessment

The patient was a housewife, and her medical history included high blood pressure for 25 years, type 2 diabetes mellitus, chronic kidney disease (CKD), osteoporosis, arthrosis, and cholecystectomy. She was assuming pantoprazole, ticlopidine, atenolol, nitroglycerin, doxazosin, simvastatin, metformin, alendronate, and non-steroidal anti-inflammatory drugs. At admission, she complained dysphagia and oral burning (glossodynia). The patient reported that the difficulty in swallowing started 1 month prior hospitalization, and progressively increased so that nourishment and fluid intake were impeded. No oral or pharyngeal lesions were appreciated at the examination. Despite pain, her face showed paucity of expression suggesting scleroderma, but Raynaud's phenomenon, skin induration, or teleangiectasia was not detected.

Serial TnI tests were normal, and a transthoracic echocardiogram showed normal ejection fraction with no regional wall motion abnormalities. Serum urea (14.30 mmol/L) and creatinine (172 μmol/L) levels (eGFR 24 mL/min/1.73 m^2 calculated with CKD-EPI formula) after an early worsening, improved with intravenous hydration. Because of bacteriuria and fever, antibiotic treatment was started, with remission of fever in 2 days.

Since the epigastric pain was associated with low levels of hemoglobin (110 g/L; red blood cells 3.63×10^{12} cells/L) and iron (8.5 μmol/L), an esophagogastroduodenoscopy was performed, which disclosed only few microerosions of the antral mucosa that could not explain dysphagia and glossodynia. To investigate whether oral symptoms were caused by

Editor: Michal Kuten-Shorrer.
Received: June 1, 2015; revised: June 21, 2015; accepted: June 22, 2015.
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The authors have no conflicts of interest to disclose.

ISSN: 0025-7974
DOI: 10.1097/MD.0000000000001163

TABLE 1. Conditions Associated With Central Sensitivity Syndrome

Fibromyalgia
Chronic fatigue syndrome
Chronic pelvic pain and endometriosis
Tension headache and migraine
Temporo-mandibular joint disorder
Idiopathic low back pain
Interstitial cystitis
Irritable bowel syndrome
Myofascial pain syndrome
Post-traumatic stress disorder
Primary dysmenorrhea
Restless leg syndrome

micronutrient deficiency, we measured plasma levels of zinc (13.0 μmol/L) and vitamin B12 (481 ng/L), but they both were normal. Because ACE-inhibitors are a well-known cause of dysgeusia, we also investigated whether the patient assumed such drugs in last years, but she denied.

Pain was then complained at the pelvis and bilaterally at the inferior limbs. Aldolase and CPK levels were normal, and an X-ray revealed osteoarthrosis.

Odynophagia and glossodynia persisted, and became the predominant symptoms. Because the patient rejected water and food for days, parenteral nutrition was started. In the next days, she also complained dysgeusia and abnormal sensitivity to some odors with nausea. Poor short-term memory was noticed, and the patient often complained to be confused ''as being in the clouds.''

After exclusion of cardiac disease and upper digestive tract lesions, the diagnosis of fibromyalgia was put forward. We therefore calculated the widespread pain index (WPI) as recommended by the American College of Rheumatology (ACR).[2] WPI of ≥7 plus symptom severity (SS) scale score of ≥5, or

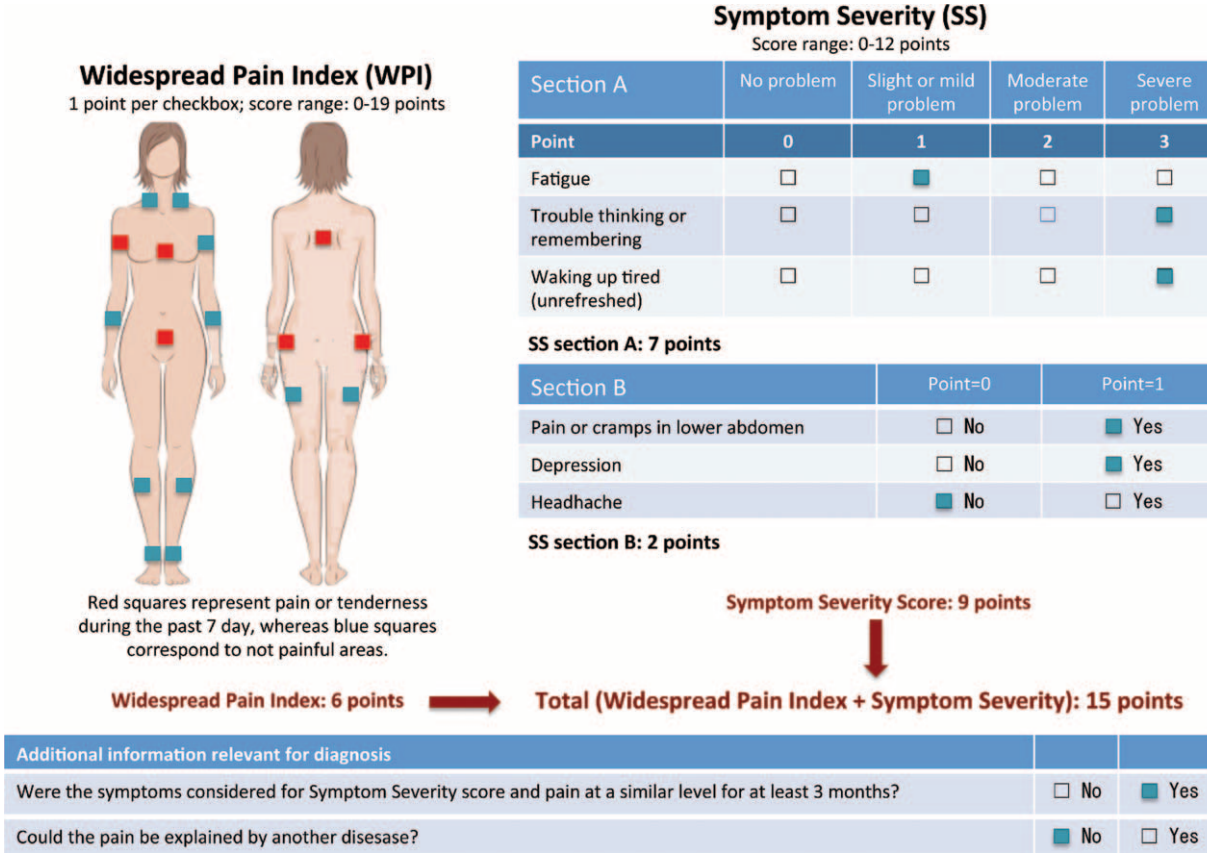

FIGURE 1. Self-report survey for the assessment of fibromyalgia in our patient.[2,3] The American College of Rheumatology recommends to assess pain and severity of symptoms using the Widespread Pain Index (WPI) and Symptom Severity (SS) Scale, which both exploit a self-report questionnaire. WPI requires that the patient localizes the sites of pain or tenderness during the past 7 days, and then the physician assigns 1 point to each site. The score ranges from 0 to 19. In our patient, the red boxes correspond to painful sites, whereas the blue to painless areas, with a score of 6. The SS Scale consists of 2 sections. In the first, that is, section A, the severity during the past 7 days is indicated for each symptom by the following scale: No problem (point 0); Slight problem: generally mild or intermittent (point 1); Moderate problem: considerable problems, often present and/or at a moderate level (points 2); Severe problem: continuous, life-disturbing problems (points 3). In the second section the symptoms listed in section B, if present during the past 6 months, are indicated by ''Yes'' (corresponding to point 1). In our patient the score was 9. The total score, which is calculated as the sum of WPI and SS, and ranges from 0 to 31 points, when ≥13 points is consistent with diagnosis of fibromyalgia. Additional informations are included in the survey (bottom) because they can be helpful for diagnosis, but no points are assigned to them. The total score of self-report survey was 15 in our patient, thereby supporting fibromyalgia (Modified from Clauw D.J. JAMA 2014).[4]

alternatively, WPI of 3 to 6 and SS scale score of $\geq$9 are the 2 criteria required for diagnosis of fibromyalgia.[2] WPI, calculated by summing up patient's reports of pain in 19 separate regions of the body as recommended by ACR guidelines, was 6 in our patient, and SS scale score, calculated by grading pain, fatigue, and ''awaking unrefreshed'' on a severity scale from 0 (no problem) to 3 (severe, pervasive, continuous, life-disturbing problems), was 9 (Figure 1).[3,4]

The final diagnosis was fibromyalgia, and treatment with amitriptyline 10 mg bid was started. No adverse events were observed.

Both odynophagia and glossodynia improved within some days, and diffuse pain, dysgeusia, and fibro-fog (ie, feeling of being in the clouds) gradually weakened in 2 weeks. At that time, the levels of hemoglobin (114 g/L) and iron (9.5 μmol/L) persisted low and, therefore, we could exclude that oral symptoms were caused by anemia.

Ethical consideration

The patient gave her informed consent to diagnostic procedures, therapy, and data collection at the admission to our Unit. She was not explicitly asked for the consent to the present case report because no element allows her identification.

DISCUSSION

Clinical presentation of fibromyalgia includes pain associated with an array of other symptoms, for example, fatigue, sleep disturbance, headache, or morning stiffness. The paucity of objective findings in contrast to the abundance of subjective ailments makes the diagnosis difficult. However, when an unusual symptom dominates the picture, the diagnosis becomes even more challenging.

Pain in fibromyalgia is widespread, entailing jaws, shoulders, arms, buttocks, legs, back, chest and abdomen, and usually is protean, being described as throbbing, stabbing, burning, cramping, or gnawing. ACR criteria require that pain is present at a similar level for at least 3 months, and that no disorder would otherwise explain it. In our 80-year-old patient presenting with chest pain, ischemic disease was excluded, and pain at pelvis and bilaterally at the inferior limbs was at a first glance referred to osteoarthrosis. The association of glossodynia and odynophagia with dysgeusia, sensitivity to odors, and fibro-fog finally unveiled fibromyalgia.

A search of the literature revealed that patients with fibromyalgia occasionally complain oral-esophageal pain and hypersensitivity. Orofacial symptoms include xerostomia, ulcerations, orofacial pain, temporomandibular joint dysfunction, glossodynia, dysphagia, and dysgeusia. Such symptoms have been seldom investigated in fibromyalgia patients, and therefore rarely reported: in a small study of 67 fibromyalgia patients who were systematically examined for orofacial symptoms, dysphagia was present in 37.3%, glossodynia in 32.8%, and dysgeusia in 34.2%.[5] Fibromyalgia was also found to be commonly associated with esophageal motility disorders,[6] nausea, and/or dyspepsia.[7,8] Hence, orofacial symptoms are more common than believed in fibromyalgia. Of interest, in our patient, odynophagia and glossodynia were the major symptoms, and their severity impeded oral nutrition, thus requiring parenteral nutrition.

We decided to use amitriptyline in our patient because depression and anxiety involve about half of the patients with fibromyalgia, and use of antidepressants is strongly advised.[9,10] Gabapentinoids, serotonin norepinephrine reuptake inhibitors, and γ-hydroxybutyrate were also found to be effective in patients with fibromyalgia.[4] In contrast, because of the hyperactivity of the endogenous opioid system in such patients,[11] opioids may lead to paroxysmal hyperalgesia.[12] The marked relief was reported by our patient after amitriptyline agrees with such reports. After 7-month follow-up, the patient is in good health with no major illness development.

In conclusion, odynophagia, dysphagia, and glossodynia can be the prevalent symptoms at presentation of fibromyalgia patients although often overlooked. The recognition of these symptoms could be determinant for a physician to pursue the diagnosis of fibromyalgia with this presentation, helpful to offer relieve of pain and avoid unnecessary tests for patients affected.

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