Restless mouth syndrome

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A 60-year-old man was referred to our headache clinic for evaluation of atypical facial pain. He reported a 1-year history of oral discomfort that began 2 to 3 months after a fall with trauma to the posterior neck. He described an uncomfortable but nonpainful sensation of swelling and numbness at the roof of the mouth that gradually spread to the tongue, bilateral buccal surfaces, and eventually the whole mouth. This was particularly prominent when his mouth was in a relaxed open position and was associated with a strong urge to move his jaw. Movement of the jaw and tongue provided temporary relief. He chewed gum for hours at a time to alleviate the symptoms, although the relief was sometimes only partial or intermittent. Keeping the jaw completely closed or clenching the teeth also provided improvement in his symptoms. Thus, he used a headband, pillow, or hand to keep the jaw closed. His symptoms began midmorning and persisted throughout the day, but were worse in the evening. The symptoms did not interfere with sleep as long as he was able to keep his jaw closed with a pillow. He denied xerostomia or xerophthalmia. He had a history of basal cell carcinoma of the left cheek status post resection 2 to 3 years ago without residual numbness. He had no personal or family history of trigeminal neuralgia, headaches, neuropathy, or restless legs syndrome (RLS). He had no history of having been on neuroleptics, antiemetics, or other dopamine antagonists. Before seeing us, he tried abstaining from caffeine and spicy food, and tried cinnamon, increased hydration, and vitamin B₁₂ without benefit. Trials of gabapentin 200 to 300 mg 3 times a day and pregabalin 200 mg twice a day also had not provided improvement. Oxcarbazepine, at an unknown dose, was ineffective and associated with anxiety and nervousness.

Physical examination, including general and neurologic examination, was normal. There was no dystonic posturing observed in the tongue at rest, during speaking, or with rapid alternating movements. Laboratory tests, including CBC, ESR, AST, ALT, TSH, B₁₂, ACE, serum protein electrophoresis with immunofixation, ANA, RF, and MPO, PR3, CCP, SS-A, SS-B, Sm, RNP, Scl 70, and Jo 1 antibodies, were either normal or negative. Ferritin was 96 µg/L. MRI of the brain showed nonspecific white matter T2 hyperintensities. MRI of the face with trigeminal protocol was unremarkable. EMG of the face including blink testing was unremarkable. EEG was negative for epileptiform activity.

Given the negative evaluation and the fact that the clinical history fulfilled the essential features of RLS involving only the mouth, he was started on pramipexole and gradually titrated up to 0.125 mg 3 times a day. At the 1-month follow-up visit, he reported considerable improvement of his symptoms, including no longer feeling the need to chew gum. At the 5-month follow-up, he reported sustained benefit on pramipexole 0.125 mg in the morning, 0.25 mg in the afternoon, and 0.125 mg in the evening.

DISCUSSION

When evaluating a patient with facial pain or discomfort, it is common to ask whether there is worsening with talking or eating, as part of the screening for trigeminal neuralgia. Our case suggests that a history of improvement with talking or eating is equally important, as it may

Practical Implications

Patients with mouth discomfort may rarely present with clinical features that resemble restless legs syndrome and benefit similarly from dopamine agonist therapy.

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introduce the possibility of an isolated regional anatomical variant of RLS. Our patient denied any pain, burning, or dysesthesia. However, it is of interest that there have been 12 cases reported of burning mouth syndrome (BMS) at least partially responsive to dopamine agonists.^{1–3} Classic BMS is characterized by an intraoral burning sensation and pain without an associated medical or dental etiology, and is typically not affected by mouth movement. In contrast, of the 12 reported dopaminergic-responsive cases of BMS, 11 reported worsening pain in the evening, and 10 reported improvement with eating, talking, or other mouth movement. Some of these patients had concurrent RLS.

RLS is characterized by unpleasant sensations associated with an urge to move the legs at rest or during inactivity that are worse in the evening or night and is at least partially relieved by movement.⁴ In recent years, studies have suggested that RLS may not be limited to the legs but may involve other isolated anatomical regions of the body with or without symptoms in the legs. Patients have been reported with dopaminergic-responsive RLS-type symptoms affecting the arms, genitals, abdomen, and bladder with and without concomitant classic RLS.^{5–8} Similarly, our patient presented with isolated mouth symptoms fulfilling the RLS diagnostic criteria in the absence of classic RLS and responded well to a dopaminergic medication. Restless mouth syndrome without features of classic RLS or BMS is rare. For patients reporting symptoms that resemble RLS but do not affect the lower limbs, a trial of a medication to treat RLS, such as a dopamine agonist, should be strongly considered.

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