Case Report: An Orofacial Pain Patient with Spots on the Brain—Multiple Sclerosis Versus Central Systemic Lupus Erythematosus

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Orofacial pain bridges an important gap between medicine and dentistry. This article presents the case of a man who reported preauricular pain, tinnitus, and vertigo that began after extraction of an impacted third molar and who was sent for evaluation of a possible temporomandibular joint disorder. However, he was subsequently found to have markers and imaging results consistent with recurrent and more centralized lupus and/or multiple sclerosis. J OROFAC PAIN 2012;26:240–243

Key words: lupus, multiple sclerosis, optic neuritis, temporomandibular disorders, tinnitus

In 1934, Costen described a syndrome of symptoms associated with the temporomandibular joint (TMJ) that led to ear and sinus symptoms.[1] Tinnitus, otalgia, and vertigo are frequent complaints in the temporomandibular disorder (TMD) population[2] and are more prevalent in TMD patients than in age-matched controls.[3] Tuz et al[4] demonstrated a higher incidence of otologic complaints (eg, otalgia, tinnitus, vertigo, and hearing loss) in 200 TMD patients compared with controls. Additional studies have also reported a higher frequency of otalgia, tinnitus, hearing loss, and vertigo in TMD patients.[5–11] TMD therapy has also been shown to be effective in decreasing ear symptoms.[12]

In the present case, due to the patient's ear symptoms following a dental procedure unrelated to any ear pathology, it was assumed this was a TMD presentation. However, since the patient had a history of systemic lupus erythematosus (SLE), optic neuritis, and migraines, TMD as a primary diagnosis was considered less likely. Therefore, imaging and further testing was obtained.

Case Report

A 49-year-old man was referred by an otolaryngologist to the University of California, Los Angeles (UCLA) Graduate Orofacial Pain and Dental Sleep Medicine Clinic with the complaint of possible TMD-related tinnitus and migraines. According to the patient, the tinnitus began immediately after extraction of two mandibular left molars, one of which was an impacted mandibular left third molar. The tinnitus was accompanied by hyperacusis, preauricular pain, and vertigo. His preauricular pain was worse on the left side but was not associated with jaw function. His tinnitus was originally on the left side but became bilateral. Previous hearing tests revealed decreased hearing in the left ear. Additionally, he reported problems with walking and balance.

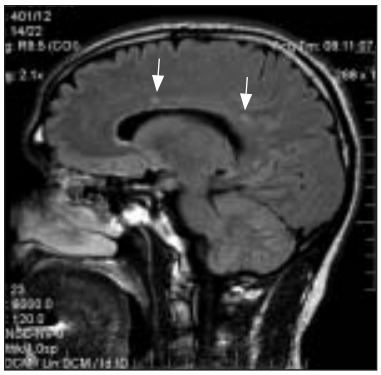

Fig 1 Sagittal MRI scan demonstrating periventricular white matter lesions *(arrows)*.

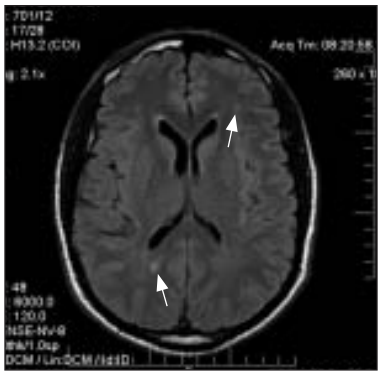

Fig 2a Axial brain MRI scan.

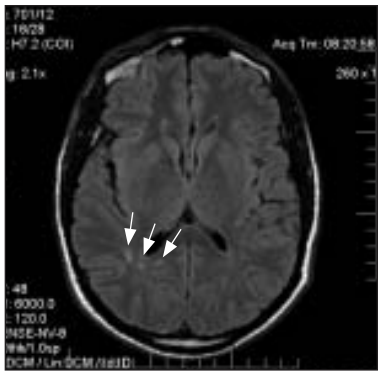

Fig 2b Additional view of axial MRI. *Arrows* indicate lesions.

Ten years prior, the patient had been diagnosed with SLE. In 2005, the patient developed optic neuritis and disturbed vision that his doctors attributed to lupus. At that time, he was given steroids and his vision improved. He had been taking Imuran (GlaxoSmithKline) and was regularly seeing his rheumatologist and ophthalmologist.

Although the patient had a long history of migraines, he began having almost daily migraines at the time of his lupus diagnosis, approximately 10 years ago. His migraines were located bilaterally in the temporal region, described as "dull and achy" with nausea, photophobia, and phonophobia and lasting for 3 to 5 hours. The migraines were preceded by auras that included visual floaters and neck pain. He was taking 0.5 mg clonazepam prn, one to two times per day to abort his headaches because of his triptan intolerance.

On examination, there were no stomatognathic findings that would indicate a TMJ etiology. His mandibular range of motion was within normal limits, with no deviation or deflection. He was missing the first through the third mandibular left molars and the mandibular right first molar, and he had moderate attrition and mild tongue/cheek ridging. His headache pain was replicated with bilateral palpation of the anterior temporalis muscles. His blood pressure was 150 over 82 mmHg and pulse rate 93 per minute. A clinical evaluation of cranial nerves II to XII revealed no abnormalities. Weber testing demonstrated increased bone conduction in the right ear, while Rinne testing was normal. The Dix-Hallpike maneuver did not produce any significant findings. However, there were positive saccades to the right with a left head thrust. Motor strength was 5/5 with no drift, and he demonstrated normal coordination and gait. Sensory tests were normal.

Brain and orbit magnetic resonance imaging (MRI) with and without contrast was performed in 2002, 2007, and 2010. The 2002 and 2007 scans demonstrated nonspecific foci of T2-weighted hyperintensity in the periatrial white matter and no abnormal orbital mass or optic nerve enhancement. However, in 2010, there were nine detected periventricular white matter lesions with subtle corpus callosal involvement, suspicious of multiple sclerosis (MS) (Figs 1 and 2). No enhancement suggesting an acute attack was apparent. Additional MRI of the cervical, thoracic, and lumbar spine was performed; none of the images showed evidence of demyelination.

Laboratory studies showed positive antinuclear antibody titers 1:80 with speckled, mitotic spindles and a strongly positive ds DNA antibody. Further cerebrospinal fluid (CSF) testing indicated elevated CSF lymphocytes and protein as well as positive antineuronal cell antibodies, in addition to high CSF IgG but negative oligoclonal bands, myelin protein. Ribosomal-P antibody was also negative.

Discussion

Inflammatory optic neuropathy is defined as any optic nerve inflammation that results in vision loss. This can be caused by demyelinating disease, sarcoidosis, orbital pseudotumor, infectious diseases such as Lyme, and rheumatologic conditions, eg, lupus.[13] It is oftentimes the first manifestation of MS, and brain MRIs frequently demonstrate white matter signal abnormalities in the absence of other clinical signs of MS.[14–18] Brain MRIs signal abnormalities at the time of the development of optic neuritis increases the probability of MS.[18–21] Following an attack of optic neuritis, there is a 20% risk of developing MS within 2 years and a 45% to 80% chance within 15 years.[22–26]

It is still unclear whether this patient's symptoms and clinical presentation are those of more central SLE or of MS. However, his CSF antibody results appear consistent with a central SLE. West et al[27] demonstrated that patients with neuropsychiatric presentations of SLE (NPLE) had abnormal CSF IgG index/oligoclonal bands, elevated antineuronal antibodies, and/or serum antiribosomal-P antibodies. This combination of tests had a sensitivity of 100%, specificity of 86%, and positive predictive value of 95%. These abnormalities also appear to lessen in NPLE patients who respond to therapy. Additional studies have also reported an increase in CSF IgG index/oligoclonal bands in NPLE patients.[28–31]

Several reports have indicated similarities between MS and SLE.[32,33] Both diseases are accompanied with high levels of viral antibodies and demyelination. A common pathogenic factor has been suggested, and strengthened by a case reported by Holmes et al[34] in which in SLE and MS occurred in homozygous twins.

Imaging

Enhancement along or within the optic nerve in optic neuritis can be revealed with an axial T-1 weighted postcontrast MRI. MS lesions are typically located around the ventricles, are ovoid shaped, and may or may not be associated with enhancement or are ring-enhancing.[13] Brain MRIs have also been useful in evaluating patients with suspected NPLE. In a study conducted by Stimmler et al,[35] 34 of 64 (53%) neuropsychiatric episodes related to SLE were associated with MRI abnormalities and were more common in patients with focal neurologic deficits and in patients with nephritis. The most frequent finding was an increased periventricular signal, although such an increase has also been described in other illnesses, aging, and MS. Diagnosis should also rely on clinical information, history, and physical examination and pertinent laboratory tests.

Conclusion

Although the exact diagnosis for this patient is still unclear, his symptoms (headaches and audiovestibular disturbances), clinical history (past diagnosis of SLE and optic neuritis), brain MRI, and laboratory testing are consistent with central and/or neuropsychiatric SLE, but the possibility of MS cannot be completely excluded. The TMD diagnosis, however, appears to be negligible as clinical examination indicated a fully functional TMJ with no musculoskeletal perpetuating factors. The patient's symptom onset and its chronological relationship with a dental extraction were most likely coincidental.

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