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Case Report

Facial myonecrosis following COVID-19[☆]Raymond Zhao, BS^a, Hadi Saab, BS^a, Anthony Propson, DO^b, Toshio Moritani, MD, PhD^a, John Kim, MD^{a,*}^aDivision of Neuroradiology, Department of Radiology, University of Michigan, 1500 E. Medical Center Dr., Ann Arbor, MI 48109, USA^bDepartment of Neurology, University of Michigan, 1500 E. Medical Center Dr., Ann Arbor, MI 48109, USA

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ABSTRACT

Myositis and myonecrosis are rare sequela of coronavirus disease 2019 (COVID-19). Until now, it has not been seen in muscles of the head and neck. We present a 22-year-old male with 4 months of retroauricular headaches following COVID-19 infection. Magnetic resonance imaging revealed rim-enhancing fluid collections in the bilateral masticator spaces which were sampled by fine-needle aspiration. We also discuss this case in the context of the current understanding of COVID-19-related myositis.

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Introduction

The COVID-19 pandemic continues to be a major problem globally. To date, various complications secondary to COVID-19 have been identified, most commonly being acute respiratory distress syndrome, acute cardiac injury, heart failure, and acute kidney injury [1]. Recently, there has been a growing body of evidence connecting COVID-19 with myositis, ranging from asymptomatic muscle enzyme elevations to severe rhabdomyolysis [2]. Herein, we present a case of bilateral masseter myonecrosis after COVID-19 infection.

Case report

A 22-year-old previously healthy male presented to our neurology clinic for chronic bilateral retroauricular tension headaches described as a 3/10 nonradiating squeezing pain with bilateral masseter muscle tightness and difficulty opening his jaw. This began shortly after a COVID-19 infection, diagnosed by PCR, 4 months prior to presentation. At the time, Omicron was the dominant strain, and he was unvaccinated. He initially also experienced severe malaise and myalgia for 4 days, which gradually resolved and did not require

Abbreviations: COVID-19, coronavirus disease 2019; MRI, magnetic resonance imaging; CT, computed tomography; DWI, diffusion-weighted imaging; ADC, apparent diffusion coefficient.

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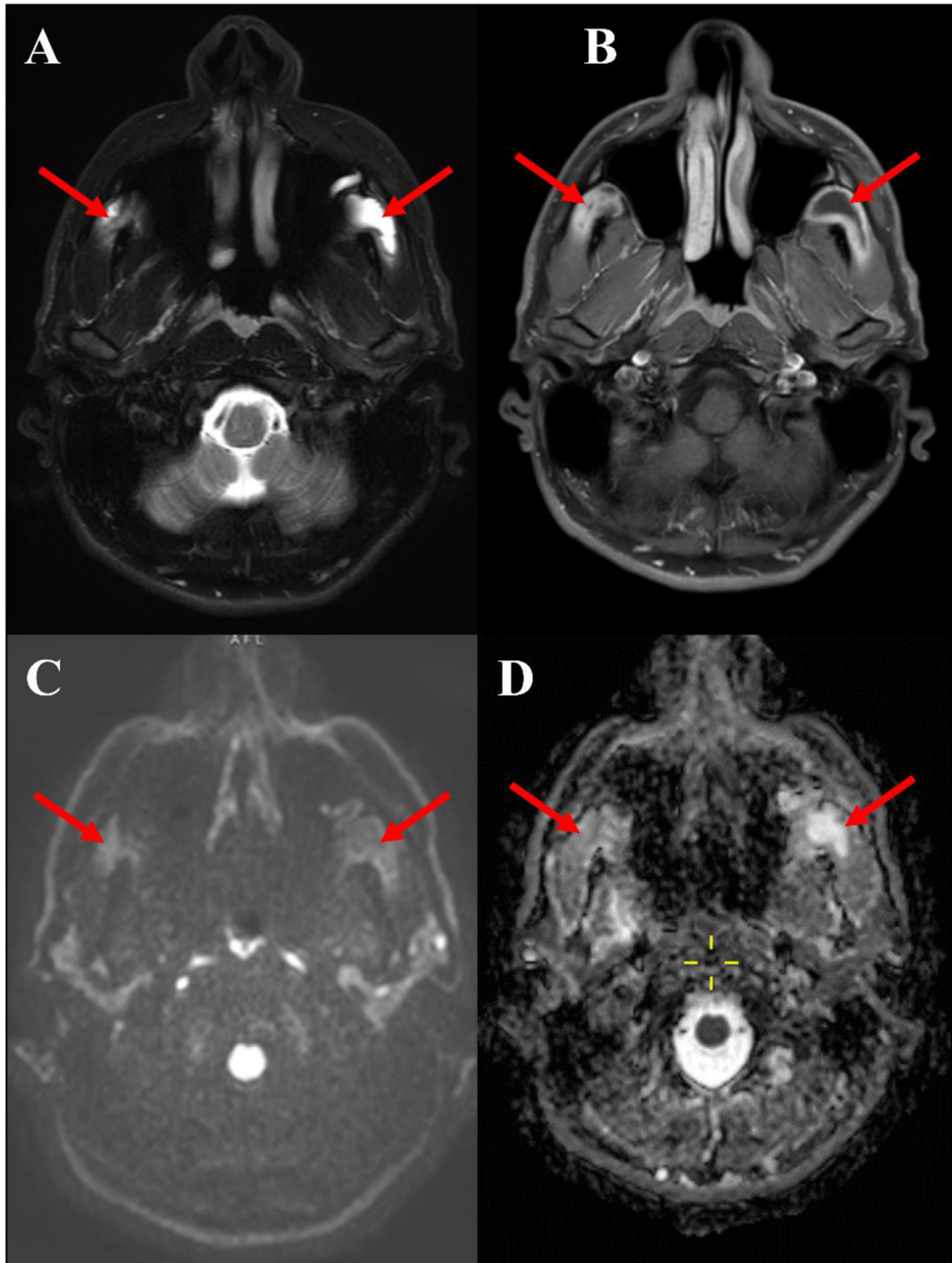


Fig. 1 – (A) T2-weighted fat-suppressed and (B) contrast-enhanced T1-weighted DIXON fat-suppressed images show rim-enhancing fluid collections (arrows) involving the anterior aspect of the bilateral masticator spaces. (C) Diffusion-weighted and corresponding (D) apparent diffusion coefficient (ADC) map shows facilitated diffusion in the center of the fluid collections.

hospitalization. However, his headaches persisted. The patient reported no neurologic or systemic abnormalities. Physical exam and laboratory evaluation were unremarkable. Previous evaluations by a local emergency room and oral surgeon were negative for temporomandibular joint (TMJ) dysfunction and meningitis.

Magnetic resonance imaging (MRI) revealed rim-enhancing fluid collections in the bilateral masticator spaces, measuring approximately 3.5 cm on the left and 1 cm on the right (Fig. 1). Diffusion-weighted imaging (DWI) and apparent diffusion coefficient (ADC) maps demonstrated facilitated diffusion in the center of the lesions (Figs. 1C and D). Due to

concern for possible infection, CT guided aspiration of the left fluid collection was performed. Bacterial and fungal cultures were negative. On cytopathology, numerous CD68 and PU.1 positive macrophages were present without malignant cells. The patient was diagnosed with myositis/myonecrosis of the bilateral masticator muscles secondary to COVID-19 and was given a 6-week prednisone course. The patient's condition has greatly improved since beginning treatment with only mild residual left sided headache.

Discussion

Myositis specifically due to COVID-19 was not formally reported until September 2020 [3]. Since then, many additional case reports have been published demonstrating various musculoskeletal manifestations including myasthenia, dermatomyositis, rhabdomyolysis, paraspinal myositis, and asymptomatic rises in creatinine kinase [2]. However, despite an increasing understanding in literature, there has not yet been a case of COVID-19-related myositis or myonecrosis presenting in the head and neck region to the best of our knowledge.

The precise mechanisms involved in the development of COVID-19-related myositis and myonecrosis is unknown. In general, there is strong evidence suggesting that autoimmune sequela of COVID-19 is due to a massive pro-inflammatory cytokine reaction, termed a “cytokine storm,” with additional contributions from hyperactivated T-cells, monocytes, macrophages, and neutrophils [4]. While these mechanisms are perhaps most associated with acute respiratory distress syndrome and multiorgan dysfunction syndrome, it's plausible that they may also underlie myositis. Specific damage to muscle tissue can occur due to abnormal activation of the adaptive immune system. SARS-CoV-2 proteins show 3 immunogenic linear epitopes that are highly similar in sequence to ones found in autoimmune dermatomyositis, suggesting similar pathophysiology through T-cell antigen presentation [5]. Stimulation of humoral immunity, cross-reactivity due to molecular mimicry, and direct-intake of viral antigens by myocytes through the ACE2 receptor are additional proposed mechanisms of muscle inflammation [2]. Lastly, a subset of myonecrosis cases may be a consequence of COVID-19's thrombotic and vasculitic milieu, possibly augmented by compartment-syndrome [6].

Diagnosis of head and neck myositis secondary to COVID-19 can be difficult given the lack of previous reports and the broad differential diagnosis for head and neck myalgia. Physicians should first rule out common or life-threatening etiologies, such as TMJ dysfunction, cluster headaches, or meningoencephalitis. After this, clinical and laboratory evaluation can provide important clues. In general, COVID-19-related myositis presents in males aged 33-88 years old [2]. While elevations in muscle enzymes are often present, they are not a reliable indicator of diagnosis or prognosis [2].

Radiographic evaluation is equally as important. In cases of COVID-19-associated rhabdomyolysis, computed tomography (CT) imaging can demonstrate hypoattenuation with or without rim-enhancement on postcontrast images [7]. When possible, MRI is the imaging modality of choice [8]. Two

patterns of imaging findings are seen on T2-weighted and contrast-enhanced T1-weighted MRI sequences: either a homogeneously or heterogeneously hyperintense region with peripheral rim of contrast enhancement [7]. In severe myonecrosis, the ring-enhancing pattern can also be accompanied by central foci of enhancement, termed the “stipple sign” [7]. Our patient presented with rim enhancing fluid collections on MRI, but no stipple sign. If a fluid collection is identified, fine needle aspiration can be performed for cytopathology to rule out nonviral infections and malignancy.

Prognosis of COVID-19-related myositis depends on the type of myopathy and its severity. In a previous retrospective review (n = 22), 21.7% of patients passed away of which 80% were due to rhabdomyolysis [2]. After diagnosis, treatment should be approached on an individualized basis. While immunosuppression can reduce inflammatory myopathies, it must be balanced with a risk of potentially worsening the viral infection itself. In a previous patient, holding mycophenolate mofetil and intravenous immunoglobulins led to reoccurrence of myositis [9]. However, in patients who are otherwise clinically stable and with low concern for re-infection such as our patient, the benefits of immunosuppression may outweigh the potential risks.

Myositis and myonecrosis are rare but important manifestations of COVID-19 that are increasingly being recognized in literature. Our case demonstrates that myalgias of the head and neck following COVID-19 infection should be carefully evaluated, especially in patients with otherwise normal clinical and laboratory examinations.

Conclusion

Myositis and myonecrosis of the head and neck region can follow COVID-19 infection. Although initial evaluation may be nonspecific, MRI and fine-needle aspiration can help confirm diagnosis.

Patient consent

Informed consent was obtained from the patient by the manuscript authors.

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