IDCases 31 (2023) e01691

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcases

Case report

Coxsackie B virus myositis in a healthy young man with mumps co-infection

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ARTICLE INFO

Keywords:
Myositis
Myopathy
Coxsackie B virus
Mumps

ABSTRACT

Infection is an established but uncommon etiology of myositis, and Coxsackie B virus has only been rarely described as a causative agent. We present a case of a 38-year-old male who presented with weakness, myalgias, and testicular pain following two weeks of upper respiratory infection. Laboratory tests revealed an elevated creatine kinase and positive serology for Coxsackie B4 and mumps. This unusual presentation of Coxsackie B myositis and mumps co-infection in a previously healthy young patient illustrates the importance of including infectious etiologies in the differential diagnosis and the potential life-threatening consequences of biased clinical reasoning.

Introduction

When faced with a patient with weakness and myalgias, the possibilities are quite broad. Even once a true muscular etiology is established, the differential diagnosis may include a number of conditions, including myopathies related to drugs and electrolyte derangements, inherited myopathies, and myositis. Myositis can be further classified into autoimmune inflammatory myositis, such as dermatomyositis, polymyositis, and inclusion body myositis, and infectious myositis due to bacteria, viruses, fungi, or parasites [1,2]. Although systemic infection commonly presents with diffuse myalgias, myositis associated with infection is less frequent, as the muscles tend to be resistant to infection. The most common etiology of viral myositis in the United States is influenza A and B, but viral myositis has also been linked to a variety of other viruses, including Coxsackieviruses [1].

Coxsackieviruses are RNA viruses in the enterovirus genus that are divided into two groups – A and B [3]. Coxsackie B infection in adults tends to present asymptomatically or with non-specific fever most of the time, but is classically associated with pleurodynia, myocarditis, and meningitis, with the possibility of severe complications such as cardiomyopathy and sudden cardiac death [4–6]. However, there have only been a few reports of rhabdomyolysis caused by Coxsackievirus [7–9]. In this report, we present an unusual case of a previously healthy young man who experienced co-infection with Coxsackievirus myositis and mumps orchitis.

Case presentation

The patient was a healthy 38-year-old male with an unremarkable past medical history who presented with diffuse muscle aches and weakness. Two weeks prior to admission, he began having a cough and sore throat; ten days after the onset of these symptoms, he woke up with muscle soreness over the entire body and weakness upon standing up from the bed. He went to urgent care, where he was prescribed prednisone 40 mg for 2 days. Over the next few days, he developed weakness, tremulousness, and numbness in his bilateral hands, rendering him unable to write or pick up his toddler son. Throughout this time, he continued to experience constant myalgias that improved with rest, as well as a sore throat with globus sensation, cough productive of light-colored sputum, and two instances of nighttime awakening with chills and night sweats. In addition, he reported pressure-like pain in the lower abdomen and edema and tenderness in the scrotum that began at the same time as the myalgias. He also noted increased urine output with progressive darkening of color since the onset of the myalgias. He had no family history of musculoskeletal disease and had not experienced trauma, intense exercise, or recreational drug usage, although his son was recently ill with hand-foot-mouth disease.

Upon admission, his temperature was 98.6 F, heart rate was 78 beats per minute, blood pressure was 148/90 mmHg, and O2 saturation was 99% on room air. Physical exam revealed a largely normal neurologic exam with patchy loss of sensation to pinprick on the forearms and

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https://doi.org/10.1016/j.idcr.2023.e01691
Received 6 December 2022; Accepted 11 January 2023
Available online 17 January 2023

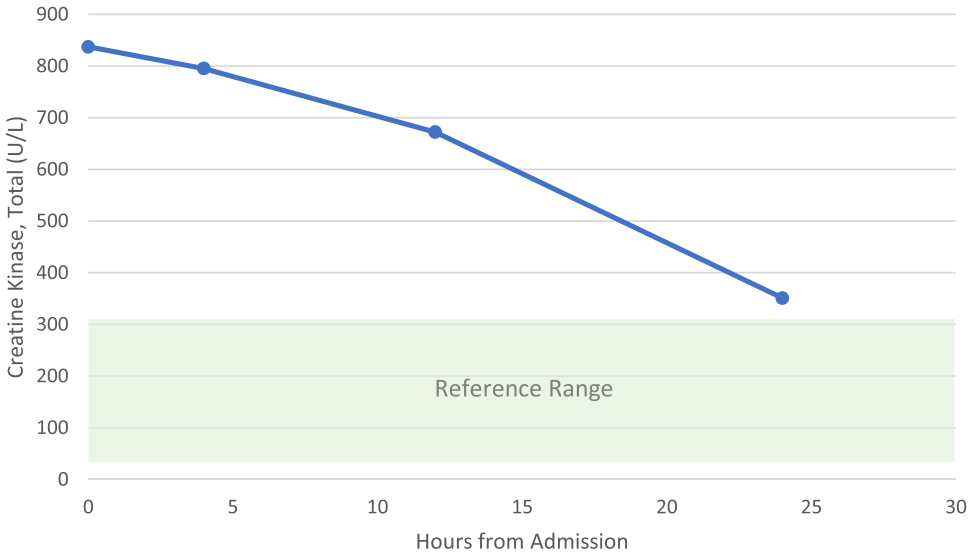

Fig. 1. Levels of serum total creatine kinase from 0 to 24 h since admission.

anterior shins. Strength was 5/5 in all extremities except for 4/5 finger abduction bilaterally and 4/5 right finger extension, and bilateral hand tremulousness was exhibited during strength testing. Diffuse lymphadenopathy was also noted. Significant initial laboratory results included an elevated total creatine kinase level of 837 U/L, elevated ALT of 71 U/L, and elevated AST of 63 U/L. No evidence of kidney injury was found. Urinalysis was unremarkable and all other biochemical and hematological values were within normal limits. Infectious and autoimmune workups were pursued. Anti-nuclear antibody was positive at a titer of 1:160, but rheumatoid factor, c-reactive protein, and erythrocyte sedimentation rate were within normal limits. An extended myositis antibody profile returned normal results. Infectious serologies were positive for Coxsackie B4 virus antibody at a titer of 1:80 and for mumps IgM but were negative for adenovirus, dengue virus, Epstein-Barr virus, cytomegalovirus, herpes simplex virus 1 and 2, influenza A and B, respiratory syncytial virus, rubella, varicella zoster, COVID-19, parainfluenza types 1, 2, and 3, and human metapneumovirus. Chest x-ray was unremarkable.

Over the next two days, the patient was treated with intravenous fluids and acetaminophen as needed for pain. Total creatine kinase levels rapidly declined to 351 U/L, ALT declined to 58 U/L, and AST declined to 46 U/L on discharge (Fig. 1). Recovery was uneventful and the patient reported gradual resolution of pain and return of muscle strength over the next month with the bilateral hands being the last to recover.

Discussion

This is an unusual presentation of infectious myositis associated with Coxsackievirus. The patient's infectious symptoms, close contact with his son who recently had hand-foot-mouth disease, and positive Coxsackie B4 antibody paint a clear picture of acute Coxsackievirus infection. Furthermore, the negative autoimmune laboratory results combined with a lack of personal and family history of myopathy suggest that the etiology of this myositis presentation is much less likely to be autoimmune and more probably infectious. While myositis is a known complication of infection, the association with Coxsackievirus is what makes this case notable. There have only been a few recorded cases of Coxsackievirus-associated rhabdomyolysis and only one other case pinpointing Coxsackie B4 as the culprit – another young adult man who experienced symptoms of upper respiratory infection before presenting with myalgias and weakness, though his creatine kinase levels were elevated to a much more severe degree than those of our patient [8]. We did not perform muscle biopsy on the patient, but it may have been able to further elucidate the underlying biological mechanisms behind such a presentation. For instance, Coxsackievirus has been isolated from the myocardium in both humans and mice, suggesting a tropism for striated muscle that one might imagine extends to skeletal muscle [6].

Complicating this case is the positive serology for mumps. Mumps is a viral infection that typically affects the pediatric population but can cause orchitis in adult men [10]. Our patient's scrotal tenderness and positive mumps IgM suggest that he had concurrent mumps orchitis at the time of admission. Of note, however, the patient had been fully vaccinated against mumps as a child, and routine vaccination has proven effective in decreasing the incidence of mumps infection [11]. Although we did not perform mumps polymerase chain reaction to confirm mumps infection, enzyme immunoassay for IgM has been found to be 99.1% specific in vaccinated patients, so it is unlikely that the result in this case was a false positive [12]. There have been reports of mumps orchitis in vaccinated male patients, and there are several theories as to why the effectiveness of mumps vaccination may decrease in certain situations [11,13]. What makes this case truly striking is the double positive serology for mumps and Coxsackievirus in a young man who whose robust baseline health and vaccination status make his rare presentation all the more unlikely.

Conclusion

This case illustrates the importance of ruling in a viral or infectious etiology when working up weakness and myalgias, especially given that Coxsackie B can lead to serious cardiac complications and chronic problems. Thorough history-taking is necessary to ensure that one does not miss a potentially life-threatening viral infection in a patient with myositis, and viral serologies may yield the diagnosis when infection is suspected. Furthermore, the finding of mumps co-infection in this patient highlights the pitfalls of making assumptions based on a patient's baseline health and vaccination status: a source of bias that carries the danger of potentially missing severe complications associated with mumps. Regardless of its influence on the treatment plan, awareness of cases such as the one described here will help direct clinicians in their medical decision making and avoid waste when similar situations arise.

Ethical approval

All study subjects partcipitated voluntarily in this study and provided their written and informed consent.

Consent

Written informed consent was obtained from the patient for publication of this casereport and accompanying images. A copy of the written consent is available forreview by the Editor-in-Chief of this journal on request.

Credit authorship contribution statement

Michelle Mao- writing. Alexander Doyle- study design.

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