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A unique case of orbital inflammatory syndrome following COVID-19 infection

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Orbital inflammatory disease is a rarely reported complication of COVID-19 infection. We report a unique case of bilateral orbital myositis in a 12-year-old boy who tested positive for COVID-19 without typical systemic symptoms. Workup for other infectious and inflammatory etiologies was negative. After failing both oral and intravenous antibiotics, the patient was started on high-dose systemic steroids, with significant clinical improvement after 24 hours, thus confirming the inflammatory etiology of his presentation.

Case Report

A 12-year-old White boy with no prior ocular history presented emergently at the Greater Baltimore Medical Center with left eye pain, eyelid swelling, and bilateral periorbital erythema of 1 week’s duration. He had already completed a course of oral cefdinir and topical moxifloxacin, prescribed by outside providers for presumed preseptal cellulitis, without improvement. He reported mild periorbital pain but was otherwise at his normal healthy baseline and denied a decrease in vision, diplopia, light sensitivity, or floaters. His corrected visual acuity was 20/25 in both eyes, with intraocular pressure of 11 mm Hg in each eye, full confrontation visual fields, and normal pupillary responses to light. Color vision was full in both eyes with Ishihara plates, and Hertel exophthalmometry confirmed no proptosis. Ocular motility testing demonstrated a −2 left supraduction deficit (Figure 1) and mild pain on abduction or adduction of either eye. External examination showed bilateral upper greater than lower eyelid edema and erythema. The remainder of the anterior segment and dilated fundus examination were normal.

Computed tomography (CT) imaging of the brain and orbits showed enlargement of multiple extraocular muscles, including the right inferior oblique muscle, right lateral rectus muscle, left superior rectus muscle, and left medial rectus muscle (Figure 2). There was also enlargement of lacrimal glands bilaterally, without signs of sinus disease or orbital abscess.

Laboratory testing, including a complete metabolic panel and complete blood count, showed a hemoglobin of 11.4 g/dL (ref, 13.5-18.0), and C-reactive protein of 3.10 mg/dL (ref, <0.5) consistent with systemic inflammation. Blood cultures were negative. Thyroid testing, ANCA screening, and ACE levels were normal. He was found to be positive for COVID-19, without classic systemic symptoms. His worsening eyelid redness and swelling despite oral antibiotics prompted admission by the internal medicine team for intravenous ampicillin-sulbactam and clindamycin. On day 3 of admission, after imaging and lab testing ruled out infection, the patient was given one dose of intravenous methylprednisolone 1 mg/kg, per ophthalmology recommendation, with significant improvement after only 24 hours. He was then discharged on oral prednisone 60 mg daily. One week after discharge, the patient’s symptoms had resolved, motility had returned to normal, and the oral prednisone was slowly tapered over 6 weeks without recurrence to date.

(See eSupplement 1, available at jaapos.org, for additional figures.)

Discussion

A thorough literature review demonstrated that the most common ophthalmic manifestations of COVID-19 have involved the anterior segment, posterior segment, and...
the neuro-ophthalmic pathway—rarely the orbit. To date, 3 reports have been published on COVID-19-related orbital inflammatory disease in children and young adults. The first included 2 separate cases of COVID-19-positive adolescents with painful unilateral eyelid swelling, and mild upper respiratory infection symptoms. CT imaging demonstrated sinusitis and fluid collections within the orbit, suggestive of orbital cellulitis, in addition to orbital or intracranial abscesses. Both patients were initially started on parenteral vancomycin and ceftriaxone, with little to no clinical improvement. The first patient was managed with an orbitotomy with drainage and subsequently demonstrated rapid clinical improvement with continued intravenous antibiotics and addition of metronidazole, nasal fluticasone, and oxyazolone topically tobramycin ointment. The second patient had near complete resolution of orbital findings after undergoing multiple endoscopic sinus drainage procedures and receiving intravenous vancomycin, ceftriaxone, and metronidazole as well as enoxaparin, hydroxychloroquine, zinc, vitamin C, thiamine, tobramycin ophthalmic ointment, and nasal fluticasone and oxymetazoline. The clinical course of both of these COVID-19 positive patients with orbital inflammation was more consistent with an infectious rather than an inflammatory etiology.

Another report discusses a 22-year-old healthy man diagnosed with right dacrtyoadenitis in the setting of COVID-19 that failed to improve with oral antibiotics but responded quickly to oral prednisone tapered slowly over 1 month. The third published report involves orbital myositis in a 10-year-old boy with periorbital swelling and magnetic resonance imaging (MRI) evidence of rectus muscle and lacrimal gland enlargement who tested positive for SARS-CoV2 in the absence of typical symptoms. This patient is most similar to the case presented here. He was prescribed oral prednisone 1 mg/kg/day tapered by 10 mg weekly and had clinical improvement within 2 days with resolution on MRI findings after 14 days.

These 4 cases demonstrate orbital inflammatory responses in young patients, all of whom had mild to asymptomatic COVID-19 infections. The literature on adult COVID-19-related orbital myositis is also limited, with the main difference being a higher prevalence of classic COVID-19 systemic symptoms, such as fever, arthralgias, or myalgias.

Orbital inflammatory disease is a rare phenomenon but has variable etiologies, including systemic inflammatory conditions, autoimmune disorders, infection and drug reactions. Various mechanisms have been proposed for COVID-19-triggered muscle inflammation. One possibility is direct viral entry via spike proteins attaching to ACE2 receptors present on muscle tissue. It is also possible that, like other viruses causing myositis, COVID-19 may activate T-cell clonal expansion and increase the production of proinflammatory cytokines, resulting in muscle inflammation and damage. Another proposed mechanism is autoimmunity due to molecular mimicry, with antibodies initially produced for host defense reacting with muscles, leading to a hyperinflammatory state with injury to myocytes. The significant response to steroids is similar to that seen in cases of idiopathic orbital inflammation (IOI). Steroid dosing for IOI, usually starting at 1 mg/kg/day of prednisone with slow taper over 6-8 weeks, can be used as a guide for treatment in COVID-19-related orbital inflammation as well.

Although we cannot rule out the possibility that COVID-19 infection was coincidental rather than causative in this case, our case report highlights the possible occurrence of orbital inflammatory disease associated with COVID-19 in the absence of typical systemic symptoms.

References
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2. Turbin RE, Wawrzusin PJ, Sakla NM, et al. Orbital cellulitis, sinusitis and intracranial abnormalities in two adolescents with COVID-19. Orbit 2020;39:305-10.
We present a case of traumatic rupture of the inferior rectus muscle associated with an inferior orbital floor fracture. Initial examination revealed a left hypertropia with severe limitation of infraduction of the left eye past the midline. The inferior rectus muscle could not be retrieved after an anterior orbitotomy. The patient underwent a variation of the modified Nishida procedure 3 months later. At 3 months’ follow-up, he had a moderate undercorrection at distance and near and a mild improvement in infraduction. He was able to fuse with prism in his glasses. Traumatic rupture of the inferior rectus muscle is an uncommon and difficult problem to treat; the variation of the modified Nishida procedure presented here offered a feasible surgical option in this case.

Case Report

A 56-year-old man with a history of hypertension and hypercholesterolemia presented with vertical binocular diplopia after blunt trauma to the left side of his face. The patient was unable to provide more detail regarding the trauma. Computed tomography of the head and orbits without contrast showed a left inferior orbital floor fracture with apparent avulsion or transection of the inferior rectus muscle (Figure 1). On presentation at Emory Eye Center the patient had uncorrected visual acuity of 20/20 in each eye. Ocular motility testing showed a left hypertropia of 25° in primary gaze, increasing to 45° in downgaze. There was poor depression in the left eye past the midline, especially in abduction (−4°). See Figure 2.

The patient underwent an exploratory orbitotomy. The cut end of the inferior rectus muscle was found posterior to the fracture site, but there was not enough viable muscle tissue to be pulled to the globe. Three months later, a variation of the modified Nishida procedure was performed as described by Nishida and colleagues,1 with the procedure performed on the left medial and lateral rectus muscles as opposed to the superior and inferior rectus muscles, as originally described. The medial and lateral rectus muscles were isolated, 6-0 polyglactin 910 suture was passed through and locked half thickness through the inferior one-third of each of the lateral and medial rectus muscles 8 mm posterior to the muscle insertion. Each suture was then passed partial-thickness through sclera 10 mm posterior to the limbus, approximately halfway between the left inferior rectus muscle stump and the horizontal rectus muscles.

One month postoperatively, the patient still reported diplopia in primary position. Examination showed an exotropia of 10° and left hypotropia of 6° in primary gaze. At near, measured in slight downgaze (reading position), he had a left hypertropia of 16°. Motility testing showed −3 infraction deficit, explaining the left hypotropia in primary gaze that switched to a left hypertropia in a reading position. Three months postoperatively, the patient reported diplopia in primary position that he could eliminate with a chin-down head position. Alignment testing showed an exotropia of 10° and a left hypertropia of 8° in primary gaze. At near in preferred reading position, he had an exotropia of 10° and a left hypertropia of 12° (Figure 3). He was able to fuse with 2° base-in prism in each eye and 4° base-up in the right eye and 4° base-down in the left eye. The patient had no follow-up beyond 3 months.

Discussion

The traumatic rupture or “snapped” inferior rectus muscle has been reported in the literature,2,3 and various transposition techniques to surgically correct the problem have been described. An inverse Knapp procedure with full-tendon transpositions has been used to transpose the horizontal rectus muscles inferiorly.3 Other authors have described excellent motor outcomes in a single case using a Hummelsheim-type procedure to reduce risk of anterior...