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Pediatric Case Reports

Ischemic Priapism in a 12 Year Old Patient Associated With Coronavirus Disease 2019 (COVID-19): A Case Report

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Infection with Severe Acute Respiratory Syndrome Coronavirus 2 (SARS-CoV-2) has been associated with changes in blood coagulation resulting in increased incidence of venous thromboembolic events and coagulopathy. Moreover, single cases of ischemic priapism have been reported in adult patients with SARS-CoV-2 infection. In this report, we describe the case of ischemic priapism in a 12-year-old child with recent SARS- CoV-2 infection. UROLOGY 165: 316–318, 2022. © 2022 Elsevier Inc.

Infection with the Severe Acute Respiratory Syndrome Coronavirus 2 (SARS-CoV-2) leads to several symptoms, ranging from fever, fatigue, and dry cough to acute respiratory distress syndrome (ARDS), with approximately 5% of affected patients requiring intensive care unit (ICU) management.[1] Moreover, a number of hematological abnormalities such as venous and arterial thromboembolic events have been described in patients with SARS-CoV-2 infection. As epidemiologic studies have shown, these complications do not occur only in the acute but also in the post-acute phase of infection, with a higher risk of thromboembolic events, even in patients not requiring hospitalization.[2]

In this context, rare thromboembolic events such as priapism have been reported in adult males.[3–8]

To fill this gap in knowledge, we report a case of ischemic priapism in a child with post-acute SARS CoV-2 infection, potentially induced by hyper-coagulability and hyper-viscosity associated with this infection.

We report this case in accordance with the CARE reporting guidelines checklist.

PATIENT INFORMATION

A 12-year-old male child presented at our outpatient emergency department with a painful persistent erection of the penis lasting for 24 hours at the time of presentation. On examination, the two corpora cavernosa were rigid, while the glans was flaccid, with no signs of skin changes, discoloration or tenderness.

The patient had a history of SARS-CoV-2-infection 7 weeks prior to the onset of priapism. Moreover, he had an anterior cruciate ligament injury and transverse fracture of the tibia plateau after a bicycle accident three days prior to the presentation. In this regard, there was no direct trauma to the pelvis or perineum. Neurologic examination was unremarkable. Furthermore, there was no history of any known hematologic disease.

At admission to our hospital, the SARS-CoV-2 PCR test was positive with a cycle threshold value of 34.9. Genetic subtyping of the virus was not performed. Laboratory testing including a complete blood count, white blood cell count with blood cell differential, platelet count and coagulation profile did not show anemia or other hematological abnormalities. The patient was also tested for SARS-CoV-2 antibodies. The results came back positive, showing 171.00 U/ml.

After circumferential local anesthesia with lidocaine-prilocaine 25 mg/g ointment, the right corpus cavernosum was punctured with an 18G butterfly needle. The blood gas analysis showed acidosis with a pH of 6.95, partial pressure of carbon dioxide (pCO_2) of 85.1 mmHg, partial pressure of oxygen (pO_2) of 3.1 mmHg and base excess of -12, confirming the clinical diagnosis of ischemic priapism. After aspiration of 40 ml of dark blood, bright red, oxygenated blood was aspirated and the blood gas analysis showed pH 7.40, pCO_2 40.2 mmHg and pO_2 125 mmHg. Despite aspiration and cavernosal irrigation with 0.9 %

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saline solution detumescence could not be achieved. Moreover, at this point, the procedure was no longer tolerated by the patient and, therefore, aborted. We decided to repeat the procedure under general anesthesia.

The corpora cavernosa were again punctured with an 18G needle and flushed with 0.9 % saline solution. The aspiration and irrigation did not lead to a full detumescence. Therefore, we injected phenylephrine with aliquots of 2 mg every 2 minutes up to 8 mg. Thereafter, the penis was detumescent. Thromboprophylaxis was initiated with enoxaparin 40 mg q.d. After 24 hours, the patient had recurrent priapism. The physical examination showed tumescent but not fully rigid corpora cavernosa and a soft glans penis. The patient was not complaining of any pain. We performed a color duplex ultrasound as well as an MRI of the penis and perineum to differentiate between ischemic and non-ischemic priapism and to exclude an arteriovenous fistula. Both imaging modalities showed a partial thrombosis of the corpora cavernosa, as well as early arterial staining of the rest of the corpora cavernosa. Thus, neither the ultrasound nor the MRI could differentiate between ischemic or non-ischemic priapism. In conclusion, at this point, there was no clear indication of re-intervention and we adopted conservative management by applying ice packs and compression to the perineum. This achieved satisfactory results.

After 3 days the patient presented with fully rigid and tender corpora cavernosa and discrete pain. A computed tomography angiography of the pelvis was performed. The exam did not show any anatomical abnormality or pelvic vein thrombosis. Under general anesthesia, the corpora cavernosa were punctured with an 18G needle. Light red blood was immediately aspirated. The blood gas analysis showed a pCO2 of 40.2 mmHg, pO2 of 125 mmHg and lactate of 1.2. The peripheral venous blood gas showed a pCO2 of 38.3 mmHg, pO2 of 158 mmHg and lactate of 0.7. As aspiration and irrigation did not lead to detumescence, a total of 5 mg of etilefrin was injected fractionally with aliquots of 1 mg every 2 minutes until full detumescence was achieved.

After observation for 24 hours, the patient was discharged. The patient was sent to the pediatric hematologic outpatient department for further investigations, where common hematologic causes for priapism, such as sickle cell disease, thalassemia, glucose-6-phosphate dehydrogenase deficiency, congenital dyserythropoietic anemia, acute lymphoblastic leukemia, and chronic myelogenous leukemia were ruled out. During a follow-up of 8 weeks, the patient did not complain about priapism recurrence and confirmed spontaneous physiologic erections.

DISCUSSION

Priapism is defined as a penile erection unrelated to sexual interest or stimulation which persists beyond 4 hours.[9] Untreated, ischemic priapism, accounting for more than 95% of all case,[10] eventually results in time-dependent smooth muscle necrosis, fibrosis of the corpora cavernosa and erectile dysfunction.[9]

The initial management of ischemic priapism consists of aspiration of corporal blood followed by irrigation of the corpora and ultimately instillation of α-agonists. If the initial management fails to achieve detumescence, shunting should be considered.[9]

In children, sickle cell disease is the most common cause of ischemic priapism but ischemic priapism may also be initiated by dehydration, glucose-6-phosphate dehydrogenase deficiency, congenital dyserythropoietic anemia, acute lymphoblastic leukemia, chronic myelogenous leukemia, mycoplasma pneumoniae infection, thalassemia, and Rocky Mountain spotted fever.[11]

It has been shown that patients with SARS-CoV-2-infection simultaneously present all 3 aspects of Virchow's triad and, hereby, the risk for thrombosis is increased. The first aspect of the triad, hyper-viscosity, may be related to blood cell count elevation, increased plasma viscosity, and impaired blood cell passage through capillaries. Hypercoagulability, as the second aspect of Virchow's triad, might result from the massive inflammatory response to the infection, with increased C reactive protein, procalcitonin, LDH, D dimer, fibrinogen, ferritin and antiphospholipid antibodies.[12] In autopsy studies, vasculopathy has been documented, possibly induced by the endothelial entrance of the virus using the ACE-2 receptor.[13] Furthermore, endothelial dysfunction, the third aspect, caused by cytokinin and free radicals, results in structural vessel wall damage and focal microvascular inflammation, which triggers endothelial activation and, thereby, pro-thrombotic conditions.[14] These pathophysiological mechanisms may ultimately lead to an obliteration of the small emissary veins in the subtunical space, which might result in ischemic priapism.

Several cases of ischemic priapism have been published, most of them affecting patients with severe symptoms requiring hospitalization and ICU admission. Lamamri et al. reported the first case of ischemic priapism in a 62-year-old man with COVID-19, which he developed upon ICU arrival.[3] The patient was treated by corporal aspiration. However, no functional outcomes have been published as the patient was not followed up. Another case was published by Addar et al., who reported a case of priapism lasting for almost 10 days in a 62-year-old male also admitted to the ICU. The case was treated by corporal aspiration and intracavernous phenylephrine injection. Surprisingly, this patient reported normal erections at his 3 weeks follow-up.[4] A third case of a 39-year-old man was published by Carreño et al., who treated the patient with local measures and intracavernous application of adrenaline. The patient died after 17 days in the intensive care unit.[5] Another 69-year-old man developed ischemic priapism in the intensive care unit. Complete detumescence was achieved by aspiration and phenylephrine. The patient had a prolonged course of severe ARDS and ultimately died.[6] A 67-year-old man unexpectedly developed priapism in the last days of his life in intensive care unit. Due to his rapid clinical deterioration, treatment focus

was redirected to best supportive care, hence, the priapism remained untreated.[7]

Giuliano et al. reported a case of ischemic priapism in a 34-year-old man with asymptomatic SARS-CoV-2 infection. The patient was treated unsuccessfully with cavernous blood aspiration and required several shunting procedures resulting in complete erectile dysfunction.[8] Therefore, in contrast to the reports mentioned above, he was not admitted to an intensive care unit and did not receive anesthetics such as propofol, as propofol was confirmed to have a strong association to priapism.[15]

In our case, considering the blood gas analysis retrieved intraoperatively during the second occurrence, we argue that the fact that the analysis showed a cavernosal oxygen partial pressure measurement above 125 mmHg can be explained by the high FiO2 during intubation, and therefore, ischemic priapism is not ruled out.

In our opinion, as no plausible other cause could be identified by meticulous examinations, the ischemic priapism of this 12-year old child is most likely linked to the previously described pathomechanisms induced by SARS-CoV-2-infection.

CONCLUSION

The clinical and laboratory presentation in our patient strongly suggests ischemic priapism related to SARS-CoV-2 infection. This medical emergency should be recognized by healthcare professionals and treated immediately to prevent erectile dysfunction. The underlying SARS-CoV-2 induced pathophysiological mechanisms need to be confirmed by future studies.

INFORMED CONSENT

Given by mother

AUTHOR CONTRIBUTIONS

All mentioned authors contributed to the writing, literary search, acquisition, and interpretation of data, critical revision, and finally the approval for publication.

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