To the Editors:

A healthy 8-year-old boy presented to the pediatric emergency ward because of a 2-day history of abdominal pain, followed by acute onset of rapidly progressive swelling and pain in the right testis. He had no history of trauma, dysuria or fever. He underwent emergency surgery because of suspicion of testicular torsion. The intraoperative finding was torsion of the right appendix testis.

After surgery, the patient developed fever with subsequent generalized rash and conjunctivitis and attended the pediatric ward of a tertiary hospital. There was a history of an upper respiratory tract infection in his father 1 month before his admission.

On physical examination, he appeared dehydrated. Vital signs included the following: temperature 39.5 °C, pulse rate of 120 beats/min, blood pressure 100/50 mm Hg and respiratory rate of 30/min. He had bilateral conjunctival erythema, maculopapular rash all over the trunk, abdominal distension with a generalized tenderness, normal breathing sound and a pansystolic murmur grade 2/6 in the left sternal border. The upper and lower extremities had nonpitting edema.

The site of the recent testicular surgery was clear without erythema or discharge. He had a normal cremasteric reflex. No lymphadenopathy was detected, and other parts of the physical examination were normal. Oxygen saturation was detected 99% at room air. The primary and midadmission laboratory results were summarized in Table 1.

The electrocardiogram was normal. The echocardiography revealed a moderate pericardial effusion and normal coronary arteries.

Considering the recent pandemic and noticeable elevation of inflammatory markers, nasopharyngeal swab polymerase chain reaction test and immunoglobulin assay for severe acute respiratory syndrome coronavirus (SARS-CoV) 2 were done. The nasopharyngeal swab was negative for coronavirus-2 or influenza (type A and B) RNA. The SARS-CoV-IgM was negative (0.23 Au/mL), but IgG was positive (5.81 Au/mL).

Regarding the patient’s clinical presentations and laboratory findings, he was diagnosed as multisystem inflammatory syndrome in children (MIS-C), and treatment with IV immunoglobulin, aspirin and corticosteroid was initiated. After therapy, the general condition improved and was discharged after 7 days.

MIS-C is a postviral inflammatory condition and the cause of scrotal pain remains unclear.2–4 However, there was no previous report on MIS-C presenting with scrotal pain and testis appendix torsion. Our patient presented with appendix testis torsion and immediately after the surgery developed fever, gastrointestinal, cardiovascular, hematologic and mucocutaneous findings. Torsion of the appendix testis is generally a self-limiting condition and the cause of scrotal pain in half of children 7–12 years of age.5

We report this case to bring awareness of atypical presenting features of MIS-C. Considering the increased emergence of MIS-C cases and lack of proper understanding of early diagnostic clinical features and treatment, there is an urgent need for more investigation on clinical presentations, treatment and outcomes.

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TABLE 1. Lab Results of the Patient

| Lab Results:                  | Initial Lab Results | Mid Admission Lab Results | Reference Range |
|-------------------------------|---------------------|---------------------------|-----------------|
| Total leukocyte count          | 14.6 × 10^9/L       | 16.2 × 10^9/L             | 4–10 × 10^9/L   |
| Hemoglobin (g/dL)              | 9.9                 | 6.6                       | 13–17 (men), 12–15 (women) |
| Platelet                       | 165 × 10^9/L        | 297 × 10^9/L              | 15–400 × 10^9/L |
| Erythrocyte sedimentation rate (mm/h) | 38              | 18                        | <5             |
| C-reactive protein (mg/L)     | 101                 | 15                        | Qualitative     |
| Blood culture                  | Negative            |                           | Qualitative     |
| Urine culture                  | Negative            |                           | Qualitative     |
| Urine analysis                 | Specific gravity 1.025 | 0.67  | 0.5–1 (children age 3–18 years) |
| Glucose 3+                      | WBC 2–4             |                           | Qualitative     |
| Stool examination              | Normal              |                           | Qualitative     |
| Na (mmol/L)                    | 130                 | 132                       | 135–145         |
| K (mmol/L)                     | 2.9                 | 3.6                       | 3.5–5          |
| Ca (mg/dL)                     | 8                   | 7.6                       | 8.6–10.3       |
| P (mg/dL)                      | 2.2                 | 3.6                       | 4–7            |
| Blood urea nitrogen (mg/dL)   | 11                  | 12                        | 10–20          |
| Creatinine (mg/dL)            | 0.66                | 0.67                      | 0.5–1 (children age 3–18 years) |
| Aspartate aminotransferase (IU/L) | 20              | 14                        | 5–40           |
| Alanine aminotransferase (IU/L) | 9                | 8                         | 7–40           |
| Alkaline phosphatase (IU/L)   | 193                 | 222                       | 44–147         |
| Total bilirubin (mg/dL)        | 0.2                 | 0.4                       | 1–1.2          |
| Direct bilirubin (mg/dL)       | 0.1                 | 0.1                       | <0.3           |
| Prothrombin time (s)          | 19.4                | 16.1                      | 11–13.5        |
| International normalized ratio | 1.44                | 1.19                      | 0.9–1.2        |
| Total protein(g/dL)           | 3.9                 | 4.3                       | 6–8            |
| Albumin (g/dL)                | 2.5                 | 2.5                       | 3.5–5          |
| Lactate dehydrogenase (IU/L)  | 422                 | 588                       | 50–150         |
| Fibrinogen (mg/dL)            | 314                 | 180–400                    |
| Troponin I titer (mg/L)       | 135.7               | 4.8                       | <19            |
| D-Dimer (mg/mL)               | 2300                | 350                       | <500           |

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To the best of our knowledge, there are only a few reported cases of coronavirus disease 2019 infection presenting with testicular pain.2,3 However, there was no previous report on MIS-C presenting with scrotal pain and testis appendix torsion. Our patient presented with appendix testis torsion and immediately after the surgery developed fever, gastrointestinal, cardiovascular, hematologic and mucocutaneous findings. Torsion of the appendix testis is generally a self-limiting condition and the cause of scrotal pain in half of children 7–12 years of age.5

We report this case to bring awareness of atypical presenting features of MIS-C. Considering the increased emergence of MIS-C cases and lack of proper understanding of early diagnostic clinical features and treatment, there is an urgent need for more investigation on clinical presentations, treatment and outcomes.

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The study is appealing but raises concerns.

A second limitation is that there was no discussion about the pathophysiological implications of the test result. Because GBS is an immunologic and not an infectious disorder, it is rather unlikely that presence of the virus in the CSF had a direct pathophysiological consequence. Anyhow, presence of SARS-CoV-2 is not uncommon. Particularly in patients experiencing meningitis or encephalitis, SARS-CoV-2 has been repeatedly found in the CSF.2–4 In immunemediated complications of SARS-CoV-2, however, SARS-CoV-2 is usually absent in the CSF. In a study of 220 patients with SARS-CoV-2–associated GBS, collected until the end of December 2020, CSF was investigated for the virus in 56 cases but was found in none of them.5 Absence of the virus in the CSF was explained by the assumption that the virus never enters the CSF or that it enters the CSF but remains only for a short time before invading neurons or endothelial cells. An argument for the temporary presence of the virus in the CSF is that virus RNA has been found on autopsy studies in neurons and endothelial cells of the frontal lobe.6

There is also no discussion via which pathway the virus had entered the CSF. Speculations in the literature include retrograde migration of the virus along cranial or peripheral nerves, hematogenic spread, or intracellular transport in leukocytes via the blood–brain barrier. Missing are the results of the cerebral magnetic resonance imaging with contrast medium. Because GBS can manifest as Bickerstaff encephalitis, it is crucial to know if there was immune encephalitis of the brainstem or not. In this respect, it should be mentioned if there was involvement of cranial nerves, the respiratory muscles or the bulbar muscles. Because GBS may be complicated by autoimmune involvement, we should know if the patient ever developed autonomic dysfunction. Although SARS-CoV-2–associated GBS is more prevalent in adults compared with children or adolescents, there is increasing evidence that also younger ages can be affected. In the study of 220 patients with SARS-CoV-2–associated GBS, 6 patients were below age 18 years.6 As a shortcoming of Table 1 is that no reference limits were provided.

Overall, the interesting study has limitations which challenge the results and their interpretation. There is a need to address these limitations to strengthen the conclusions.

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Letters to the Editor

Presence of SARS-CoV-2 in the CSF of Guillain-Barré Syndrome Patients Requires Validation

To the Editors:

We appreciate the interest in our article1 and the opportunity to respond to the comments. In the published report, we present a pediatric case of COVID-19-associated GBS