The First Report of *Sphingomonas yanoikuyae* as a Human Pathogen in a Child With a Central Nervous System Infection

To The Editors:

*Sphingomonas* spp. are aerobic, yellow-pigmented, glucose nonfermenting, Gram-negative bacteria and a rare cause of human infections.1 Herein, we report a case of *Sphingomonas yanoikuyae* central nervous system infection in a child. This is the first report of *S. yanoikuyae* as a human pathogen to the best of our knowledge.

A 31-month-old male with a history of type 3 von Willebrand disease, decompressive craniectomy due to intracranial hemorrhage and tracheostomy was admitted to the hospital with fever and swelling in the skin flap area. The computed tomography revealed hydrocephalus, and a shunt operation was planned via neurosurgery. Before surgery, magnetic resonance imaging was performed and revealed a collection compatible with subdural empyema with an approximate diameter of 3.5 cm. Therefore, abscess drainage and external ventricular drain placement were performed. Cerebral spinal fluid (CSF), blood and abscess cultures were negative. On the 58th day of hospitalization, he had a fever, and *S. yanoikuyae* was isolated from the CSF culture. It was sensitive to meropenem, tobramycin and amikacin. Treatment was switched to meropenem. However, his CSF cultures grew recurrent *S. yanoikuyae*, and intrathecal amikacin was added to his treatment. After 14 days of meropenem and 5 days of intrathecal amikacin treatment, his CSF culture became sterile. Total meropenem therapy was discontinued after 28 days.

In conclusion, our case is the first report of *S. yanoikuyae* as a human pathogen. Due to this specimen’s limited antimicrobial susceptibility data, it is challenging for clinicians to treat this rare bacteria. We recommend raising awareness of this rare organism associated with nosocomial infection, especially in patients with indwelling intravascular devices.

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REFERENCES

1. Yabuuchi E, Yano I, Oyaizu H, et al. Proposals of *Sphingomonas paucimobilis* gen. nov. and comb. nov., *Sphingomonas parapaucimobilis* sp. nov., *Sphingomonas yanoikuyae* sp. nov., *Sphingomonas adhaesiva* sp. nov., *Sphingomonas capsulata* comb. nov., and two new species of the genus *Sphingomonas*. *Microbiol Immunol*. 1990;34:99–119.

2. Lin JN, Lai CH, Chen YH, et al. *Sphingomonas paucimobilis* bacteremia in humans: 16 case reports and a literature review. *J Microbiol Infect Dis*. 2010;43:35–42.

3. Bayram N, Devrim I, Apa H, et al. *Sphingomonas paucimobilis* infections in children: 24 case reports. *Mediter J Hematol Infect Dis*. 2013;5:e2013040.

4. Tai ML, Velayuthan RD. *Sphingomonas paucimobilis*: an unusual cause of meningitis-case report. *Neural Med Chir (Tokyo)*. 2014;54:337–340.

5. Vaz-Moreira I, Nunes OC, Manaia CM. Diversity and antibiotic resistance patterns of *Sphingomonadaceae* isolates from drinking water. *Appl Environ Microbiol*. 2011;77:5697–5706.

Crimean-Congo Hemorrhagic Fever Mimicking Multisystem Inflammatory Syndrome in Children Associated With COVID-19: A Diagnostic Challenge

To the Editors:

Crimean-Congo hemorrhagic fever (CCHF), which is endemic in our country, is a zoonotic disease characterized by fever and hemorrhage that may be severe and even fatal. The primary transmission route to humans is tick bite; however, other forms of transmission are also reported including direct contact with blood or other bodily fluids of infected animals, nosocomial and vertical transmission.1 The clinical diagnosis of CCHF is difficult to establish because symptoms are nonspecific in the prehemorrhagic phase of the disease and laboratory findings are similar to other childhood infectious diseases.2,3 Here, we report two cases...
with fever, elevated inflammatory markers, and multisystem organ involvement initially considered as multisystem inflammatory syndrome in children (MIS-C), but subsequently diagnosed with CCHF—highlighting the diagnostic challenge in distinguishing MIS-C from other infectious diseases. To our knowledge, no CCHF cases misdiagnosed as MIS-C have been reported to date.

Case 1

A 9-year-old boy was referred to our clinic with a preliminary diagnosis of MIS-C because of persisting fever, abdominal pain, conjunctivitis and increasing levels of acute phase reactants despite intravenous antibiotic treatment. His mother had been diagnosed with coronavirus disease-2019 (COVID-19) 2 months prior to admission. On admission, he had tachycardia (130 beats/min), conjunctivitis and maculopapular rash. His laboratory examination revealed lymphocytopenia, thrombocytopenia and elevated levels of liver function tests, troponin, pro-brain natriuretic peptide (pro-BNP) and inflammatory markers including procalcitonin (Table 1). Although all MIS-C criteria were met, CCHF diagnosis was also suspected, despite lack of tick bite, since the patient had progressively decreasing platelet count, and had a history of living in an endemic area and his family was engaged in animal husbandry. Polymerase chain reaction (PCR) test for CCHF was positive, and the patient was treated and discharged without any complications.

Case 2

A 15-year-old boy was referred to our hospital with a preliminary diagnosis of MIS-C because of having fever, conjunctivitis, gastrointestinal symptoms, elevated inflammatory markers and history of exposure to his father who had documented COVID-19 infection three months ago. On admission, he had fever, bilateral conjunctivitis, and malar rash. Laboratory analysis revealed lymphocytopenia and thrombocytopenia in addition to elevated inflammatory markers (Table 1). Detailed questioning revealed that he had a history of close contact with animals; however, history of tick bite was not present. On the following days, his fever persisted and platelet count continued to decrease. CCHF PCR was positive and he was treated successfully.

To conclude, similar to MIS-C, clinical manifestations of CCHF include fever, headache, myalgia, rash, conjunctivitis, abdominal pain, nausea, vomiting and diarrhea. Detailed patient history and differential diagnosis are important to avoid unnecessary treatment and protect healthcare providers since infection control measures are essential in CCHF. Our two cases highlight that CCHF should be ruled out when evaluating possible MIS-C, even in the absence of tick bite history, especially in patients with progressive decline in platelet count, those living in endemic areas, and those with a history of close contact with animals.

**REFERENCES**

1. Tezer H, Polat M. Diagnosis of Crimean-Congo hemorrhagic fever. *Expert Rev Anti Infect Ther.* 2015;13:555–566.
2. Kara SS, Kara D, Fettah A. Various clinical conditions can mimic Crimean-Congo hemorrhagic fever in pediatric patients in endemic regions. *J Infect Public Health.* 2016;9:626–632.
3. Centers for Disease Control and Prevention. CDCHAN-00452, May 14, 2020. Multisystem inflammatory syndrome in children (MIS-C) associated with coronavirus disease 2019 (COVID-19). 2020. Available at: https://emergency.cdc.gov/han/han00452.asp. Accessed May 28, 2021.

**TABLE 1. Clinical and Laboratory Findings of Patients**

| Organ system involvement | Case 1 | Case 2 |
|--------------------------|--------|--------|
| Cardiovascular            | ✓      | No     |
| Respiratory               | No     | No     |
| Gastrointestinal          | ✓      | ✓      |
| Hematologic               | ✓      | ✓      |
| Renal                     | No     | No     |
| Neurologic                | No     | No     |
| Dermatologic              | ✓      | ✓      |

| Organ system involvement | Case 1 | Case 2 |
|--------------------------|--------|--------|
| SARS-CoV-2 RT-PCR        | Negative| Negative|
| SARS-CoV-2 antibody      | Negative| Negative|
| History of COVID-19 exposure | Yes | Yes |
| CCHF-related characteristics | ✓      | ✓      |
| History of tick bite     | No     | No     |
| History of close contact with animals | Yes | Yes |
| CCHF PCR                 | Positive| Positive|
| Treatment                | Ribavirin| Ribavirin|

ALT indicates alanine aminotransferase; AST, aspartate aminotransferase; BNP, brain natriuretic peptide; CCHF, Crimean-Congo hemorrhagic fever; COVID-19, coronavirus disease 19; CRP, C-reactive protein; LDH, lactate dehydrogenase; MIS-C, multisystem inflammatory syndrome; RT-PCR, reverse transcriptase polymerase chain reaction; SARS-CoV-2, severe acute respiratory system coronavirus 2.