

# Recurrent Intussusception as a Manifestation of COVID-19

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<b>Background</b>	The COVID-19 pandemic has resulted in over 288,000 known childhood cases. In recent literature, there have been scattered cases of intussusception as a manifestation of COVID-19, including two infants that progressed to intestinal necrosis and septic shock.
<b>Summary</b>	We present a case of intussusception with concurrent COVID-19 infection complicated by multiple episodes of early recurrence after complete pneumatic reduction.
<b>Conclusion</b>	This case, along with others previously published in the literature, suggests intussusception as a possible manifestation of a COVID-19 and highlights the potential danger of presuming those with COVID-19 harbor the same low risk of intestinal necrosis and intussusception recurrence.
<b>Key Words</b>	COVID-19; severe acute respiratory syndrome coronavirus 2; SARS-CoV-2; intussusception, intussusception recurrence, air-contrast enema

**DISCLOSURE STATEMENT:**

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## Case Description

The coronavirus disease 2019 (COVID-19) pandemic caused by acute respiratory syndrome coronavirus 2 (SARS-CoV-2) is a major public health crisis that has resulted in over 288,000 known childhood cases with a 0.3 percent mortality rate in the pediatric age group.<sup>1</sup> This worldwide pandemic began in December 2019, when a novel coronavirus was discovered in Wuhan, China, after a series of severe pneumonia cases of unknown etiology.<sup>2-4</sup> The virus quickly spread around the globe despite efforts to decline its spread. With the outbreak reaching its peak, there has been an associated increase in reported cases of infected children and infants.<sup>1</sup> Common symptoms include the expected fever and dry cough; however, gastrointestinal symptoms have also been frequently seen.<sup>5,6</sup>

There have been scattered case reports of intussusception in children as a manifestation of COVID-19 within the literature, including two children who progressed to intestinal necrosis, septic shock, and multiorgan failure resulting in their death.<sup>7-9</sup> We present a case of intussusception as the presenting manifestation of a COVID-19 infection, complicated by multiple episodes of early recurrence after complete pneumatic reduction.

The patient is a healthy six-month-old male who presented to the emergency department with a four-day history of episodic abdominal pain with associated increased irritability, emesis, and passage of bloody mucus per rectum. His mother also reported he had experienced occasional unmeasured fever the past few days, but he was afebrile on presentation and had no noted respiratory symptoms. His mother had been tested for COVID-19 at another hospital the day prior due to subjective fever and mild respiratory symptoms; however, her results were unknown at the time of his presentation. The surgical team caring for the patient was unaware that she had test results for COVID-19 pending. An abdominal ultrasound was performed consistent with an ileocolic intussusception, and he was taken to the fluoroscopy suite with successful pneumatic reduction of the intussusception. Per protocol at the institution, he was observed overnight for possible recurrence. Approximately 12 hours after the successful reduction, he had recurrent abdominal pain and irritability with evidence of recurrence on ultrasound. Pneumatic reduction was again performed, with successful complete reduction on the second attempt. A few hours later, the child's mother was notified she had tested positive for COVID-19, and a nasopharyngeal swab for COVID-19 was performed. He continued to have poor oral intake and irritability, so another ultra-

sound was performed approximately six hours after his second successful reduction, which demonstrated that he had re-intussuscepted for a third time, again successfully reduced with an air-contrast enema. After this reduction, his COVID-19 test results came back positive. He was monitored an additional 17 hours without recurrence of symptoms prior to discharge home. Throughout his hospitalization, he remained afebrile without evidence of additional COVID-19 symptom development.

## Discussion

Beginning as a few cases of pneumonia with unknown etiology in Wuhan, China, the novel coronavirus, coined COVID-19, has developed into a worldwide pandemic that has rapidly spread around the globe.<sup>4</sup> Thankfully, in the pediatric population, most experience an asymptomatic or mild course, and deaths remain extremely rare.<sup>10</sup> In the largest pediatric cohort to date, Dong et al. noted critical illness with COVID-19, exhibiting ARDS or organ failure, to only be present in 0.4 percent of children.<sup>11</sup> Although the most common reported symptoms are fever and dry cough, present in 43 to 44 percent of cases, gastrointestinal (GI) manifestations including diarrhea, abdominal discomfort, and vomiting are also frequently seen, reported in 2 to 18 percent of patients.<sup>2,5,6,12</sup> In a study of adult cases of COVID-19, Wang et al. noted approximately 10 percent of COVID-19 patients presented with GI complaints, often seen one to two days before the development of fever and respiratory manifestations.<sup>13</sup> Similarly, in a previous case series of hospitalized children positive for COVID-19 without respiratory symptoms at presentation, 80 percent were found to have GI symptoms as the first manifestation of the virus, including gastroenteritis, acute suppurative appendicitis, and a case of intussusception. Although these children did not exhibit respiratory symptoms, some exhibited impaired liver and renal function, and chest computed tomography (CT) images in all cases revealed ground-glass opacities and consolidation consistent with pulmonary COVID-19 involvement. Unfortunately, in the case of intussusception reported in this series, the child progressed to sepsis and multiorgan system dysfunction with necrosis of the proximal ileum requiring exploratory laparotomy and resection; after which he passed away of multiple organ failure.<sup>7</sup> A similar case was reported by Lu et al., a 10-month old with intussusception who tested positive for COVID-19, also resulting in intestinal necrosis, septic shock, and multiorgan failure, dying four weeks after admission.<sup>5</sup> Contrastingly, another published case of intussusception with associated COVID-19 is a six-month-old boy who presented with classic symptoms

of intussusception without respiratory symptoms or fever. In this case, the intussusception was successfully reduced with hydrostatic reduction, and he was subsequently discharged home without known recurrence and presumed full recovery.<sup>9</sup>

Diagnosis of the virus is performed by reverse transcriptase-polymerase chain reaction (RT-PCR) on a respiratory specimen via nasopharyngeal or oropharyngeal swab.<sup>14</sup> The sensitivity and specificity of these tests remain largely unknown. Available data suggests very high specificity (around 98 to 99 percent) with only moderate sensitivity (approximately 63 to 78 percent), making a positive test to likely indicate the presence of COVID-19 but a negative test inadequate to rule out infection, especially in the presence of known exposure or symptoms.<sup>15</sup> Although not used for diagnosis, live viral ribonucleic acid has been repeatedly detected in stool samples of infected patients, further indicating involvement of the gastrointestinal tract, with a noted longer clearance of viral particles in feces than seen in the pharynx.<sup>12,14,16</sup>

It remains unclear why children and young adults appear less severely affected than their older counterparts, and the true pathogenesis of the virus is not yet fully understood. It is suspected to be due to a combination of differences in immune system function as well as a variation of expression of the angiotensin-converting enzyme 2 receptor (ACE2) through which the virus infects.<sup>6</sup> This receptor is abundantly present in lung alveolar epithelial cells, leading to the typical respiratory signs as dry cough and dyspnea of COVID-19; however, it is also highly expressed in enterocytes of the small intestine and colon. The gastrointestinal manifestations seen in COVID-19 infection, as well as the dissemination seen in some cases, including involvement of vascular endothelium, myocardium, liver, kidney, skin, and central nervous system, is likely due to local ACE2 expression. It is known ACE2 expression in the lungs increases with age, providing a possible factor in the decreased severity of pulmonary manifestations within the pediatric age group.<sup>16</sup> The possibility also exists initial gastrointestinal manifestations are due to direct infection of the GI tract by fecal-oral transmission, and appropriate contact precautions should be followed; however, this is not yet fully elucidated in the literature.<sup>17</sup>

Increased prevalence of intussusception in the setting of a viral illness involving the gastrointestinal tract is not unexpected. Precedent infections such as adenovirus and rotavirus have been associated with intussusception, and approximately 30 percent of intussusception cases have an

antecedent viral illness before onset, likely through hypertrophy of the Peyer's patches of the intestine resulting in a propulsive lead point.<sup>18</sup> COVID-19 enterocyte involvement likely increases the risk of intussusception through the same mechanism, although further studies evaluating intussusception in COVID-19 are needed. Regardless, previously reported cases highlight the importance of a heightened suspicion for possible COVID-19 infection in the setting of intussusception, especially in the presence of fever or possible exposure.

Treatment of intussusception includes hydrostatic reduction, pneumatic reduction, as well as operative reduction. In the pediatric population, unless there is evidence of ischemic necrosis or a pathologic lead point necessitating surgery, the initial treatment of choice is hydrostatic or pneumatic reduction under fluoroscopic or ultrasound guidance. The success rate of pneumatic reduction ranges from 83 to 89 percent;<sup>19–20</sup> however, with a risk of recurrence of 1 to 13 percent.<sup>21–23</sup> Studies have found increased recurrence risk in children over two years old, with a duration of symptoms less than 12 hours or greater than 48 hours, rectal bleeding, left-sided intussusception, and those with a noted pathologic lead point.<sup>21,24–25</sup>

The presented case is unique in that the infant did not progress to ischemic necrosis and multiorgan system failure as seen in previously reported cases of intussusception with concurrent COVID-19; however, he experienced repeated early recurrence, requiring pneumatic reduction three times within a 20-hour period. Due to a risk of recurrence, historically, infants with intussusception have been monitored for some time after hydrostatic or pneumatic reduction. However, recently there has been a push within the literature for early discharge without routine admission, citing a low risk of recurrence within the first 48 hours.<sup>19,22,24</sup> Reported experience of intussusception in the setting of COVID-19 is still limited at this time; nonetheless, this case and other published cases of complicated intussusception in COVID-19 positive children suggest those with concurrent COVID-19 infection may not have the same low risk of recurrence and complications. Therefore, a period of observation may be warranted. In addition, as many COVID-19 patients present without the classic respiratory symptoms and febrile illness, it is also important to consider that intussusception may be the presenting sign of an infection and a negative COVID-19 status should not be assumed. As we continue to observe and study this novel virus, the impact of a concurrent COVID-19 infection and intussusception will hopefully be further elucidated.

## Conclusion

The presented case is notable in several ways. It highlights the potential error of assuming children who present with the appearance of idiopathic intussusception do not have an associated COVID-19 infection, even without any pulmonary manifestations or fever. Additionally, it demonstrates the danger of presuming those with concurrent COVID-19 infection have the same low risk of early intussusception recurrence, as this patient exhibited multiple episodes of recurrence within a 20-hour period of observation. As evidenced by previously published cases, these patients are also at a significantly higher risk for progression to bowel ischemia and necrosis, even death. Due to the increased risk, we suggest monitoring these patients for an observational period may be prudent and prevent unnecessary morbidity and possible mortality.

## Lessons Learned

This case, along with others previously published in the literature, suggests intussusception as a possible manifestation of a COVID-19 and highlights the potential danger of presuming those with COVID-19 harbor the same low risk of intestinal necrosis and intussusception recurrence.

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