CASE REPORT

Unique Oral and Behavioral Manifestations of Post-COVID Multisystem Inflammatory Syndrome in a 5-year-old Child: A Rare Case Report

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Abstract

Background: Multisystemic inflammatory syndrome (MIS) in the post-coronavirus disease (COVID) scenario is recognized in the pediatric population worldwide. However, no case report features jaw bone necrosis caused by self-mutilating injury noted during COVID treatment.

Case description: This report is of a 5-year-old child affected by COVID. During the course of treatment, she presented with MIS and was treated with steroids and oxygen therapy. She was observed to have cardiac and gastrointestinal disorders. However, some psychiatric impairment caused her to pull out her lower anterior teeth, thus acting as an injury to the tissue and causing osteomyelitis. This necrotic bone tissue was excised surgically, and due to the loss of teeth, prosthetic rehabilitation was performed for the patient.

Conclusion: This unique presentation of post-COVID manifestations of self-mutilating injury is alarming for the pediatric population.

Keywords: Case report, Coronavirus disease 2019, Osteomyelitis, Self-mutilating injuries, Steroids.

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Introduction

In December 2019, Wuhan, China, received the initial report of the novel coronavirus disease 2019 (COVID-19), which is caused by severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2). The World Health Organization (WHO) declared the COVID-19 outbreak a public health emergency on January 31, 2020.[1] Approximately, 456 million people worldwide have been impacted by this public health disaster as of March 2022, and more than six million people have died as a result.[2] Children with acute COVID-19 have a relatively reduced hospitalization and mortality rate compared to adults.[3] Several names were used to characterize the condition at the beginning of the pandemic, including pediatric inflammatory multisystem syndrome temporally associated with SARS-CoV-2 (PIMS-TS) and multisystem inflammatory illness in children and adolescents. The illness has been called multisystem inflammatory syndrome in children (MIS-C) linked with COVID-19 after both the Centers for Disease Control and Prevention (CDC) and the WHO produced case definitions.[4] Respiratory failure, cardiac dysfunction, hematological crises, and gastrointestinal symptoms are a few of the different MIS-C presentations.[5] The exact prevalence of oral abnormalities in MIS-C and the clinical and prognostic relevance of these findings are unknown. Frontline healthcare professionals might have trouble identifying these subtle but potentially significant changes, in contrast to dentists and other oral healthcare experts who are used to documenting oral mucosal diseases. There are various cases reported in the literature that describe the oral manifestations of erythematous mucous membranes and tongue with or without aphthous stomatitis.[6] The present case is unique, presenting with eccentric symptoms of hallucinations, aggressive behavior, and self-mutilating injuries, such as pulling out primary mandibular anterior teeth, leading to osteomyelitis in conjunction with positive COVID-19 testing.

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Patient consent statement: The author(s) have obtained written informed consent from the patient's parents/legal guardians for publication of the case report details and related images.

Case Description

A 5-year-old patient reported to the department of pediatric and preventive dentistry with a chief complaint of a hard mass in the lower front teeth region. Clinical examination revealed exposed bone in the mandibular anterior teeth region, extending from the primary mandibular left canine to the primary mandibular right first molar. The medical history revealed a persistent fever 1 month prior, for which she was admitted to a private hospital. Routine blood tests indicated an infectious etiology, and typhoid, dengue, and malaria tests were negative. As the patient was unstable, a two-dimensional (2D) echocardiography test was done, which was normal. Later on, COVID swabs tested positive using

reverse transcription polymerase chain reaction (RT-PCR). Due to economic constraints, the patient was shifted to a government hospital after 1 week.

Thereafter, the patient received oxygen and steroid therapy: methylprednisolone (250 mg) once a day and injection dexamethasone (0.6 mL) every 8 hours for 5 days. Due to the administration of steroid therapy, she developed generalized edema (Fig. 1). Further routine blood investigations revealed anemia and thrombocytopenia. The second echocardiography revealed myocarditis with mitral and tricuspid valve regurgitation. Thereafter, the patient received aspirin (75 mg and 150 mg on alternate days). She also received 5 units of platelets and fresh frozen plasma for 3 days.

During this period, the patient complained of repeated episodes of nausea, vomiting, abdominal pain, dermatitis, hair exfoliation, psychotic and aggressive behavior toward her mother, and a hallucinatory episode in which she saw cars crashing into her body. She also pulled out all her primary mandibular anterior teeth. After 18 days of hospitalization, she was discharged and advised to continue taking aspirin (75 mg) once in the morning and prednisolone (5 mg) once at night for 3 months. The patient was then referred to a dental hospital for the treatment of her teeth. Intraoral examination revealed missing primary mandibular anterior teeth and necrosed bony socket and alveolar process of the mandibular anterior region (Fig. 2). The exposed bone was not tender and firmly adhered to the lower jaw with no signs of bleeding. The orthopantomography revealed a radiolucent band over the crowns of the permanent mandibular anterior teeth (Fig. 3). Surgical excision of the necrosed bone was planned under local anesthesia. Surprisingly, the child was consuming a double dose of aspirin and prednisolone and was referred for pediatric clearance. As the prothrombin time and international normalized

Fig. 1: Overall health of the patient

Fig. 2: Intraoral examination revealed necrosed bone in mandibular anterior region

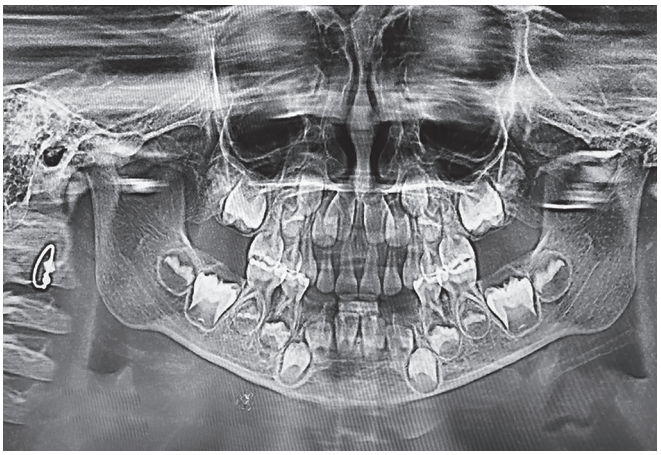

Fig. 3: Preoperative digital orthopantomogram

Fig. 4: Necrosed bone after its removal

Fig. 5: Surgical site immediately after removal of necrosed bone

Fig. 6: Healing of the site seen during recall after 30 days

Fig. 7: Eruption of permanent mandibular incisors noted during recall after 6 months

ratio were increased, the patient was advised to take vitamin K and stop steroids and aspirin 7 days before the surgery. Surgical excision of the necrosed bone (curettage and trimming of sharp bony sequestra using a bone file) from the mandibular region was performed under local anesthesia when all the blood parameters were within normal limits (Figs 4 and 5). Postoperatively, the patient was instructed to continue aspirin for 3 months, along with antibiotics and analgesics for 5 days. After 30 days, healing of the surgical site was uneventful (Fig. 6). A recall of the patient was also done 6 months after the surgery. Complete healing of the surgical site was noted, along with the eruption of permanent mandibular incisors (Fig. 7). However, due to the mobility of the incisors, a modified lingual arch appliance was planned to be given at a later date.

Discussion

The clinical spectrum of pediatric patients with laboratory-confirmed COVID-19 and MIS-C is acute and severe, with a significant mortality rate. During diagnosis, 80–97% of MIS-C patients experience abdominal pain, vomiting, and/or diarrhea.[7] This could aid in identifying this severe multisystemic illness during the ongoing pandemic. In routine clinical practice, early detection of MIS-C may be supported by the presence of serious additional characteristics, such as hypoxemia, arterial hypotension, myocarditis, pericardial effusion, and shock.[7,8] However, in the present patient, additional observations were noted, such as psychotic and aggressive behavior, hallucinations, and self-mutilation traits, such as pulling out teeth. These symptoms have not been reported in children who are COVID-19 positive to date.

The pathophysiology and mechanisms by which SARS-CoV-2 triggers an abnormal immune response leading to MIS-C remain poorly understood. Serum biomarkers that indicate inflammation, such as C-reactive protein, D-dimer, troponin, and ferritin, have been shown to be elevated in the presence of MIS-C. The poor outcome, multi-organ dysfunction syndrome, and cytokine storm have all been linked to elevated levels of these indicators.[9,10]

Multiple rounds of immunosuppressive medications were used on these individuals, who also showed significant illness complexity and severity, an increase in hospitalizations, and a higher susceptibility to severe secondary infections.[11] The current patient was treated with anti-inflammatory medications and systemic corticosteroids. Long-term steroid use may influence serotonergic systems, which could consequently increase the likelihood of depression, attention-deficit/hyperactivity disorder, impulsive behavior, and other psychiatric symptoms.[12,13] In this case report, we intend to shed light on the mental impairment caused to the child, which led to hallucinations where she thought cars were running into her and self-mutilating injuries such as pulling out her own teeth. These issues might also be related to the consumption of high doses of steroids. The act of pulling out her teeth, combined with the presence of infective foci in the body, disrupted the healing of the oral wound, leading to osteomyelitis. Furthermore, since antibiotics are unable to reach this area, surgical intervention is required. Therefore, it was decided to perform surgery to remove the necrosed bone. The patient's prosthetic rehabilitation should begin as soon as the surgery site has fully healed.

Conclusion

Few studies have summarized the oral symptoms of MIS, and it is still unknown if these lesions are due to a coronavirus infection or a result of the patient's systemic illness. Since support, pain management, and quality of life are important, it is crucial to highlight the early detection of COVID-19 oral symptoms to properly define the role of pediatric dentists.

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