Case Report

Acute submandibular sialadenitis: a possible presentation of COVID-19 in children

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Abstract

This case report describes a paediatric patient presenting with acute unilateral submandibular sialadenitis during the 2019/2020 SARS-CoV-2 pandemic. The patient's history, clinical examination and investigatory work-up favoured a viral aetiology over bacterial infections or sialolithiasis. Laboratory tests for common viral aetiologies of sialadenitis were negative. SARS-CoV-2 PCR testing was positive. Indirect evidence supports the hypothesis that SARS-CoV-2 can cause acute sialadenitis. Caregivers could consider the possibility of SARS-CoV-2 infection in an afebrile child with acute sialadenitis presenting during the current pandemic.

Introduction

Saliva has various functions, including lubrication, taste, digestion, tooth protection, antibacterial properties. It is produced by the salivary glands and transported into the mouth by the salivary ducts. Viral infections are the most common acute salivary gland disorders in children (1).

Our patient presented in a Belgian hospital during the 2019-2020 severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) pandemic. SARS-CoV-2 is a human pathogenic betacoronavirus causing coronavirus-disease 2019 (COVID-19). There is a wide array of possible clinical presentations, ranging from asymptomatic carriers to severe bilateral pneumonia with acute respiratory distress syndrome to isolated anosmia. In general, children appear to run a milder clinical course.

Case report

A 10-year-old boy presented at the emergency department with an acute onset painful swelling of the right submandibular area. Moreover, he was feeling markedly unwell for the last two days. He reported difficulties eating and speaking. He did not have any tooth pain. He reported a recent history of having high fever (>39°C), a dry cough and breathing difficulties during the previous week, which spontaneously resolved. During the last four days, respiratory symptoms and fever were absent. The patient received all of his recommended vaccines, had no history of similar episodes and had no significant medical history. Familial history revealed that both parents had been sick recently, both reporting dry cough and fever. The mother had a negative polymerase chain reaction (PCR) test for SARS-CoV-2. Clinical examination showed an ill-defined area of swelling of the right submandibular region (see Figure 1). The area was slightly red, warm and tender on palpation. The

Figure 1. Clinical picture showing residual swelling of the right submandibular area, taken two days after initial presentation.

patient displayed trismus; he was unable to open his mouth more than 2 cm. He was visibly dehydrated with dry mouth mucosa and a prolonged capillary refill time. Oral examination revealed no signs of tooth decay and normal appearance of the salivary duct orifices. Lung auscultation was normal. Laboratory investigations showed slightly elevated inflammatory markers, with C-reactive protein (CRP) levels of 26 mg/L (<1 mg/L), leukocytosis of 11 x 10^9/L (4-10 x 10^9/L) with mild neutrophilia of 8,54 x 10^9/L (2-8 x 10^9/L) and lymphopenia of 1,42 x 10^9/L (3-9 x 10^9/L). Ultrasound revealed inflammation of the right submandibular salivary gland and reactive swollen cervical lymph nodes, without sialolithiasis or signs of abcedation. The patient was admitted for intravenous fluid resuscitation, pain medication and intravenous antibiotics (amoxicillin/clavulanic acid). Serologic testing for cytomegalovirus (CMV), Epstein-Barr virus (EBV) and mumps virus was negative. PCR testing for SARS-CoV-2 on nasopharyngeal swab was positive. During the course of hospitalisation, the patient did not display symptoms or signs suggestive of pneumonia caused by SARS-CoV-2. The swelling decreased gradually over the following days.

Discussion

Our patient presented with unilateral acute submandibular sialadenitis. In children, sialadenitis is most frequently caused by viral infections. Bacterial sialadenitis and sialolithiasis were also considered in the differential diagnosis. The absence of fever, the absence of pus on clinical examination and only slightly elevated inflammatory parameters on blood tests rendered bacterial infection unlikely. Nevertheless, our patient received intravenous antibiotic treatment to account for the possibility of a bacterial infection. Sialolithiasis is a rare cause of sialadenitis in children. Ultrasound is regarded as the first imaging modality of choice for detecting sialoliths and detects up to 90% of sialoliths greater than 2 mm (2). Ultrasound imaging did not show any sialoliths in our patient, which rendered sialolithiasis unlikely. Testing for viral organisms known to frequently cause sialadenitis (including mumps virus, EBV and CMV) was negative. A full respiratory screening panel was not performed. PCR testing for SARS-CoV-2 was performed because of the current global pandemic and was positive.

Various viruses have been reported to cause sialadenitis. In the pre-vaccine era, the mumps virus was the most frequent cause of sialadenitis in children. Since the start of widespread vaccinations against the mumps virus, various studies investigated other potential viral aetiologies of sialadenitis using PCR-based analysis or serologic testing. The most frequently found viral aetiologies of mumps negative sialadenitis are EBV, influenza, parainfluenza and human herpesviruses. Barrabeig et al. performed the only study that included human coronaviruses (coronavirus 229E and coronavirus OC43) in their PCR-based analysis (3). They investigated 101 suspected mumps cases with negative PCR results for the mumps virus and reported no positive results for these two coronaviruses in their study population (3).

A recent study in Medical Hypotheses by Wang et. al investigated whether SARS-CoV-2 has the potential to cause acute (and/or chronic) sialadenitis (4). SARS-CoV-2 invades host cells by binding to the angiotensin-converting enzyme 2 (ACE2) receptor. This receptor is present in salivary gland epithelium, with a mean expression level of the ACE2 gene being even higher in salivary gland tissue than in lung tissue (5). The virus can indeed be detected in salivary specimens, with a high viral load in saliva, comparable to those in sputum and throath swabs (6). Wang et. al hypothesised that SARS-CoV-2 can bind to ACE2 receptors on salivary gland epithelium, fuse with them, replicate and induce cell lysis causing acute sialadenitis (4). Moreover, excessive immunoreaction may further damage the salivary glands, similar to tissue damage seen in other organs with ACE2 receptor expression. Secondary fibrosis could later lead to chronic sialadenitis (4). They concluded that the indirect evidence indicates a high probability that SARS-CoV-2 does have the potential to cause acute (and chronic) sialadenitis (4). There are, however, no studies that systematically investigated SARS-CoV-2 infection in patients presenting with acute sialadenitis during the current pandemic. So far, there are only three reported cases of acute sialadenitis caused by SARS-CoV-2 (7, 8). They include two adult patients with unilateral parotitis and one adult patient with combined bilateral submandibular sialadenitis and parotitis.

Conclusion

This article is the first case report on acute submandibular sialadenitis in a SARS-CoV-2 positive paediatric patient. Indirect evidence supports the hypothesis that SARS-CoV-2 can cause acute sialadenitis. Caregivers could consider the possibility of SARS-CoV-2 infection in an afebrile child with acute sialadenitis presenting during the current pandemic. More evidence is needed to establish a definite link between SARS-CoV-2 infection and acute sialadenitis.

The authors declare that there is no conflict of interest.

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