The restrictive effects of the COVID-19 pandemic on the management of plastron appendicitis in a known hemophilia A patient

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ABSTRACT

The coronavirus disease-2019 (COVID-19) pandemic has caused many people to fear and worry about getting infected. The atmosphere of anxiety and restrictions due to the pandemic has caused challenges in the diagnosis and treatment of diseases. In this case report, we aim to present the effects of the COVID-19 pandemic on the delay in the diagnosis and treatment of plastron appendicitis in a patient with hemophilia A. A 21-year-old male patient with hemophilia A was admitted to our emergency department in April 2020 with complaints of abdominal pain and vomiting for 4 days. His computed tomography findings were compatible with plastron appendicitis with a diameter of 61x36 mm in the right lower quadrant. The patient was interned for general surgery. After 7 days of antibiotic therapy, the patient was discharged with oral antibiotics. The patient was called for a follow-up appointment 10 days later, and the operation was planned for 6 weeks later. The patient did not attend the follow-up appointment due to government lockdown measures and his fear of the pandemic. The patient was admitted to our emergency department with complaints of right groin pain and discharge in the right groin in May 2020. The patient stated that he stopped taking antibiotics 7 days after discharge, failing to complete his treatment course. On the computed tomography of the patient, it was observed that the plastron abscess opened into the right rectus sheath and fistulated to the skin. The abscess was drained, and the patient was re-admitted to the hospital for intravenous antibiotic therapy. The patient underwent elective appendectomy after factor VIII replacement in June 2020. The patient was discharged on the 7th postoperative day. In conclusion, uncertainties due to the pandemic environment, fear of the coronavirus pandemic, and problems in drug supply caused disruption and delay in his treatment process. Even in the presence of a pandemic that can cause abdominal complaints such as COVID-19, acute appendicitis should always be considered in the preliminary diagnosis of abdominal pain in young patients with a diagnosis of hemophilia A.

Keywords: Plastron appendicitis, COVID-19, pandemic, hemophilia A

INTRODUCTION

Coronavirus disease-2019 (COVID-19) first appeared in December 2019 in Wuhan, China, as a disease caused by severe acute respiratory syndrome coronavirus-2 (¹). The disease has spread rapidly and turned into a worldwide public health crisis (¹). As a result of the increased public health risks, the outbreak was declared a pandemic by the World Health Organization on 11 March 2020 (²). The COVID-19 pandemic has caused many people to fear and worry about being infected (²). As a result of this, a reduction in emergency department admissions for non-COVID-19 low severity acute medical conditions has been observed (³). Appendicitis is a medical condition that needs urgent surgical treatment (⁴). Acute appendicitis occurs when the appendix lumen is blocked due to various reasons, which could be due to feces, lymphoid hyperplasia, infections, and rarely a tumor obstructing the appendix lumen (⁵). For the clinical diagnosis of appendicitis, the Alvarado scoring system.
is used, which categorizes patients as low-intermediate and high risk depending on signs, symptoms, and laboratory findings consistent with appendicitis including: the presence of right lower quadrant tenderness, elevated temperature, rebound tenderness, migration of pain to the right lower abdomen, anorexia, nausea and vomiting, leukocytosis and leukocyte left shift (5). No biomarker is specific alone in the diagnosis of appendicitis. Among the imaging methods, computed tomography (CT) is the most widely used imaging method, however, in the pediatric age group, transabdominal ultrasonography is preferred to avoid radiation exposure (5). Appendicitis is divided into simple and complicated. Complicated appendicitis has clinical reflections such as gangrenous perforation and abscess formation (5). Plastron appendicitis is described as the formation of an abscess surrounded by the omentum due to complicated perforated appendicitis (6). Plastron appendicitis can present with abdominal pain, nausea, vomiting, diarrhea, and an abdominal mass (6). According to a meta-analysis that included 20 studies with 59,448 patients, the incidence of plastron appendicitis in adults is 4.8%, more commonly seen in males (7, 8). Ultrasonography and CT scans have high specificity and sensitivity in the diagnosis of this condition. Treatment of plastron appendicitis is surgery followed by conservative treatment (7). These gastrointestinal symptoms like abdominal pain, nausea, vomiting, and diarrhea are also seen in COVID-19 (9). This situation may cause diseases that require urgent surgical treatment, such as appendicitis, to be overlooked (2). Emergence of the COVID-19 pandemic has delayed hospital admissions, making it difficult for patients to get diagnosed (10). The pandemic has also caused difficulties in supplying medicines and personal protective equipment (11). These implications can change disease management, delay the diagnosis and cause medical cases to be more complicated (3, 10).

Hemophilia is an inherited bleeding disorder in which the blood does not clot properly (12). Hemophilia A is a coagulation disorder caused by factor VIII deficiency. Hemophilia A is X-linked recessive and is diagnosed in approximately 1 in 5000 male births worldwide (13). The most common complaint of hemophilia A patients is bleeding in the musculoskeletal system, which can include mucous membranes (13, 14). It is crucial to determine whether symptoms such as fever and vomiting that occur with abdominal pain in hemophilia patients are caused by hemophilia or due to another disease (13). In addition to COVID-19, the presence of additional diseases that cause coagulation disorders such as hemophilia A may complicate the diagnosis and treatment process of diseases, like appendicitis in this case (15).

In this case report, we aim to present the effects of the COVID-19 pandemic on delaying the diagnosis and treatment of plastron appendicitis in a patient with hemophilia A. Informed consent was obtained verbally.

## CASE REPORT

A 21-year-old male patient with known hemophilia A was admitted to our emergency department in April 2020 with abdominal pain and vomiting for 4 days. On examination, there was tenderness in the right lower quadrant with guarding and rebound tenderness with a negative indirect rebound. The patient's leukocyte count was 15000/uL and C-reactive protein (CRP) was 6.52 mg/dL. From clinical findings, the patient's Alvarado score was calculated to be 7/10. In his CT there was an appearance compatible with plastron appendicitis with a diameter of 61x36 mm in the right lower quadrant. The patient was interned to general surgery. The patient was started on intravenous (IV) antibiotics (Ceftriaxone 1x2000 mg IV). After 7 days of antibiotic therapy, the patient was discharged with oral antibiotics. The patient was scheduled for a follow-up appointment in 10 days, and his surgery was planned for 6 weeks later. The patient did not attend his follow-up appointment 10 days later due to government coronavirus measures. Five weeks later, he presented with pain and discharge from his right groin. He stated that he stopped taking antibiotics 7 days after discharge. The patient's leukocyte count was 13400/uL and CRP was 0.6 mg/dL. In his CT scan, it was seen that the plastron abscess had opened into the right rectus sheath and fistulated into the skin. After factor VIII replacement therapy, abscess drainage was performed under general anesthesia. The patient was followed up with Amoxicillin + Sulbactam 4x1.5 g IV and Ornidazole 2x500 mg IV antibiotics for 11 days, alongside a wet dressing. The patient was discharged with an oral prescription for Amoxicillin + Clavulanate 2x1000 mg and Ornidazole 2x500 mg. The patient was called for a follow-up appointment 15 days later. In the follow-up CT, the patient's plastron findings had strongly regressed. It was decided to continue oral antibiotics for 15 days and then operate. In June 2020, the patient was hospitalized for elective surgery. After factor VIII replacement therapy, an open appendectomy was performed. The patient's abdominal drain was removed on the fifth postoperative day, and the patient was discharged on the seventh postoperative day.

## DISCUSSION

During the COVID-19 pandemic, elective surgeries have been postponed in many countries, and most hospitals have been reserved to serve only emergency and COVID-19-related cases (16). There are various methods of protection from coronavirus spread such as vaccination, masks, room ventilation, and maintaining social distance (17). For some people, social distancing and fear of COVID transmission may delay seeking medical attention for other medical conditions (16). In addition, policies implemented by authorities, such as handling nonserious diseases at home may have been a reason for the decrease in hospital admissions (16). Because of the pandemic, country-wide lockdowns were declared in many countries.
During the lockdown period, a reduction in hospital admissions and hospitalizations, including emergency departments, was noted (16, 18). In addition, it has been reported that there has been a decrease in cases requiring emergency surgery, like acute cholecystitis and appendicitis (simple and complicated) (16). However, despite the decrease in the number of admissions of complicated appendicitis, the rate of admission for surgery has increased compared to the pre-pandemic period (16). As a result, the pandemic has hindered patients requiring emergency surgery from attending the hospital due to the fear of being infected with COVID-19 and has caused these patients to present in a more complicated fashion later on (19).

The treatment of plastron appendicitis is still controversial. While conservative treatment with antibiotics is appropriate for simple plastron appendicitis, percutaneous or surgical drainage is more appropriate for plastron appendicitis with periappendicular abscesses (7). According to Andersson et al. (8), for conservative treatment, the recurrence rate is 7.2% and the failure rate is 7.6%. The risk of malignancy from appendicitis treated non-surgically is 1.2%, and it has an increased cancer rate for those over 40 years of age if not surgically treated (7).

Appendectomy, which is one of the most frequently performed operations in general surgery, cannot be performed in some delayed presentations due to plastron formation and is performed as an interval appendectomy three months later, as it was found that the morbidity rate decreases as appendectomy is delayed (20). Most surgeons prefer open appendectomy for these cases due to possible adhesions (20).

In addition to these, some factors can complicate the diagnosis, hemophilia being one of them (13). Diagnosis is difficult in patients with hemophilia presenting with abdominal pain, as bleeding should be considered first in the differential diagnosis of hemophilia patients presenting with acute abdominal pain. However, diagnoses such as appendicitis and peptic ulcer perforation may also be the differential diagnoses and coexist with hemophilia A (13). Hemophilia A commonly presents with hemorrhage into the skin, muscles, soft tissues, and mucous membranes (14). Consequently developed symptoms like nausea, abdominal pain, fever, leukocytosis, and vomiting can also be seen in acute appendicitis (21, 22). Therefore, the diagnosis of acute appendicitis with the Alvarado score in a patient with hemophilia A may not be reliable enough. CT is important in such patients in order not to miss appendicitis and other possible causes of acute abdomen (13).

There are two main approaches to replacement therapy in the treatment of hemophilia: First, prophylaxis to prevent bleeding events, and second, management to treat bleeding when it occurs (13). Recombinant factor VIII is crucial in perioperative patient care, as inhibitory antibodies to factor VIII may develop. The response to factor VIII replacement is predicted by the presence of inhibitory antibodies (15). If the inhibitor level is high, plasmapheresis can be used to lower the inhibitory antibody level before elective surgery, but in patients requiring emergency surgery, plasmapheresis is neither useful nor practical for reducing antibody levels (15). As a surgical approach, factor VIII should be increased above 80% just before the operation to ensure blood clotting cascade during surgery, and dynamic monitoring is essential before, during, and after surgery (13). In our case report, the patient was given 2000U factor XIII twice, on the morning of the operation and two hours before the operation. Postoperatively at the sixth hour, the patient was again given 2000U of factor XIII.

In conclusion, the social isolation conditions required by the pandemic negatively affected individuals with hemophilia in terms of disease management and drug supply (23). The World Federation of Hemophilia has issued guidelines on the need to delay non-urgent medical care and elective surgery and minimize hospital visits for patients with hemophilia who contract COVID-19 (23). As in our case report, with the addition of comorbidities, medical care and treatment can be delayed in such patients. The difficulty experienced by patients with hemophilia in drug supply is more pronounced in low-income countries than in high-income countries (24). However, there is insufficient clinical data on hemophilia patients with COVID-19 (23). Therefore, this case report is unique in that it includes COVID-19, hemophilia A, and plastron appendicitis. Uncertainties arising from the pandemic and fear of infection may delay diagnosis in such cases, as well as making the diagnosis and management of the disease difficult and causing problems in drug supply. Even in the presence of a virus that can cause abdominal complaints such as COVID-19, acute appendicitis should always be considered in the preliminary diagnosis of abdominal pain in young patients with a diagnosis of hemophilia A. For this reason, health professionals should consider this situation while managing the diagnosis and treatment process.

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Informed Consent: Informed consent was obtained verbally.

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