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Acute abdomen in post-Covid 19 multisystem inflammatory syndrome in children. A case report.

AIM: *As more data about coronavirus disease-2019 (COVID-19) has been gathered it has become evident that children who have had or have been exposed to COVID-19 can develop a rare complication; multisystem inflammatory syndrome in children (MIS-C). We report the case of a 9-year-old boy, who was brought to the emergency room with suspected acute abdomen and was diagnosed with MIS-C.*
METHODS: *The patient had had a positive molecular test for COVID-19, 25 days earlier and fever that started 4 days prior to presentation, He tested negative for COVID on arrival at the emergency room. After physical examination, and diagnostic tests were performed the differential diagnosis included appendiceal inflammation and MIS-C. Surgical exploration was performed laparoscopically.*
RESULTS: *The immune morphological picture was reactive lymphogranular hyperplasia. Postoperatively the abdominal symptoms improved rapidly but the patient developed diffuse erythema as well as some cardiovascular and neurological disturbances. The patient was discharged on postoperative day 14 in good general condition with a diagnosis of MIS-C.*
CONCLUSIONS: *In patients with a recent positive COVID test and mainly gastroenterological manifestations surgical exploration is necessary in order to prevent delayed diagnosis and inadequate/inappropriate treatment.*

KEY WORDS: Acute abdomen, COVID-19, MIS-C, Gastrointestinal symptoms

Introduction

Multisystem inflammatory syndrome in children (MIS-C) is a rare but potentially lethal syndrome, apparently related to Coronavirus disease 2019 (COVID-19) [1]. This syndrome has a variety of clinical manifestations and there are no pathognomonic features or definitive diagnostic tests [1,2]. The diagnostic criteria of the Royal College of Paediatrics and Child Health (RCPCH), the Centers for Disease Control and Prevention (CDC) and the World Health Organization (WHO) differ, but they all include fever, evidence of systemic inflammation and involvement of at least ≥2 organ systems [3-55].
The most common presenting symptoms are gastrointestinal and in most cases the children have fever present for ≥4 days [6,7]. Features of MIS-C may resemble those of Kawasaki disease, toxic shock syndrome and macrophage activation syndrome [1]. Several cases of MIS-C presenting with a clinical picture of acute abdomen, have been described, and some of these patients underwent surgical exploration [6,8]. We report a case of a 9-year-old boy, who tested positive for COVID, 25 days before presentation, with symptoms that suggested acute abdomen.

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Clinical Case

A 9-year-old boy was brought to the emergency room of the Taranto Hospital (Italy), with suspected acute abdomen. He had no significant medical history except for a positive molecular test for COVID-19 25 days earlier and fever that started 4 days prior to presentation, for which he was on antibiotics since 2 days. On admission he tested negative for COVID. He appeared to be severely ill complaining of abdominal pain and nausea. Abnormal findings included fever (39.5 °C) tachycardia (145 bpm), a palmar rash, and petechiae on his chest, His abdomen was painful on superficial and deep palpation with percussion tenderness especially in the lower quadrants. .Blumberg's sign and Rovsing's sign were positive. There was marked elevation of biomarkers of inflammation (C-reactive protein, procalcitonin) and coagulation (fibrinogen and D-dimer). Chest X-ray showed an enlargement of the upper mediastinum. Abdominal ultrasound and total body computed tomography (CT) scan revealed diffuse abdominal lymphadenopathy, dense, diffuse intraperitoneal free fluid, thickening of the sigmoid colon and signs of appendiceal inflammation.

This constellation of findings led us to perform a surgical exploration Laparoscopic exploration revealed diffuse turbid serous fluid, distension of the appendix and vascular congestion, and diffuse lymphadenopathy. The remaining macroscopic findings were normal.

Appendectomy and biopsy of omental lymph nodes was performed. Analysis showed preserved lymph node structure with marked histocytosis of the sinuses, lymphoid follicles with germinal centers mainly in the secondary phase of development. Immunohistochemical characterization (CD3; CD20; CD10, bcl2; CD5, CD23, Ki67/MIB1) demonstrated normal distribution of B and T cell zones Culture of the intra-abdominal fluid was negative.

The histological examination of the appendix revealed follicular hyperplasia and chronic lymphoplasmacellular inflammation. The immunomorphological picture was reactive lymphogranular hyperplasia.

During postoperative observation the patient developed plantar and scrotal erythema and bilateral subconjunctival hemorrhage. The levels of inflammatory markers rose. The abdominal symptoms on the other hand, progressively improved with the recovery of intestinal function on day 1.

Infusion of immunoglobulins was started and then intravenous steroid therapy with progressive clinical improvement A marked increase in pro-BNP and troponin levels was observed and the patient developed a small pericardial effusion. On postoperative day 2, the patient had visual hallucinations. The electroencephalogram showed changes compatible with encephalitic involvement but a CT scan of the brain was negative. A lumbar puncture was performed that was negative for COVID-19 (experimental research) and other viruses related to encephalitis. In 48 hours the clinical picture normalized.

The patient was discharged on postoperative day 14 in good general condition with a diagnosis of MIS-C. All symptoms had resolved.

Discussion

MIS-C is a rare but complex and potentially lethal condition that usually occurs 3-4 weeks after COVID infection, often after the patient no long tests positive for COVID. The description of MIS-C was established by the CDC on the basis of clinical and laboratory criteria as shown in (Table I) [4].

Data about the pathogenesis of MIS-C- is still limited, The clinical picture of MIS- is complex and the diagnosis and management of MIS-C patients is challenging A health advisory issued by the CDC states that "Persistent fever without a clear clinical source is the first clue. Any fever that is accompanied by symptoms concerning in their severity or coincident with recent exposure to a person with COVID-19 should raise suspicions" [4].

In a retrospective chart review of 44 children who were hospitalized with a diagnosis of MIS-C, Miller et al note that patients most commonly present with gastrointestinal manifestations (84.1%) accompanied by fever(100%) and often rash(70.5%) [9]. The clinical picture can be that of acute abdomen or atypical appendicitis [8,10,11].

On presentation our patient had predominant gastrointestinal symptoms with fever and a mild palmar rash and petechiae on his chest.

However, preoperative laboratory and instrumental findings, were not consistent with a diagnosis of a primarily abdominal pathology. Procalcitonin, C-reactive protein, fibrinogen and D-dimer levels were higher than what is commonly observed with primary surgical causes of acute abdomen. The patient also had a pericardial effusion. Moreover CT scan showed marked and widespread supra- and subdiaphragmatic lymphadenopa-

Table 1 - *CDC criteria*

All 4 criteria must be met:
1. Age <21 years
2. Clinical presentation of MIS-C, including:
   - Fever: to 24 h
   - Laboratory evidence of inflammation
   - Multisystem involvement to 2 organ systems
   - Severe illness requiring hospitalization
3. No alternative plausible diagnosis
4. Evidence of recent SAR-CoV-2 infection/exposure:
   - Positive SARS-CoV-2 (RT-PCR)
   - Positive serology
   - Positive antigen test
   - COVID-19 exposure within 4 weeks prior to the onset of symptoms

thy. The thickening of the walls of the appendix and the sigmoid colon was difficult to interpret. In the light of these findings. The differential diagnosis included MIS-C, acute bacterial peritonitis , acute appendicitis and acute onset lymphoproliferative disease.
The clinical picture observed led us, in agreement with pediatricians to opt for surgical exploration. A minimally invasive technique was adopted to optimize the risk-benefit ratio.
Laparoscopic exploration made it possible to rule out acute inflammatory processes affecting the appendix, and other abdominal organs. The presence of widespread lymphadenopathy was confirmed and it was possible to perform a biopsy of the omental lymph nodes. The patient quickly recovered gastrointestinal function, on day 1, and resumed oral intake on day 2. In the following days additional inflammation was observed, especially mucocutaneous inflammation and neuroinflammation, making the diagnosis of MIS-C clearer.
A systematic review on MIS-C by Rouva et al (385 cases) showed that almost 50 % of the children who presented with suspected acute abdomen underwent surgery. Although half were negative a surgical pathology was confirmed in almost 25% of cases [12]. We agree with the authors' observation that, currently, relying on clinical findings alone might not be sufficient in the management of this patient population.

Conclusions

Post-COVID, MIS-C is a rare syndrome characterized by a varied clinical presentation that occurs a few weeks after exposure to or infection with COVID-19. Because of the limited data available the best treatment approach cannot be determined.
We believe that in patients with a recent positive COVID test and mainly gastrointestinal manifestations surgical exploration is necessary in order to prevent delayed diagnosis and inadequate/inappropriate treatment of potentially lethal disease.

Riassunto

L'infezione da SARS COVID-2 ha caratteristicamente un interessamento respiratorio nei soggetti adulti pur dando frequentemente sintomatologia gastroenterologica.
Di recente sono state segnalate manifestazioni cliniche correlate alla infezione COVID 19 che interessano pazienti pediatrici e consistono in manifestazioni infiammatorie multisistemiche (MIS-C) talvolta analoghe alla malattia di Kawasaki. Risultano poche e sporadiche segnalazioni di MIS-C con un quadro di addome acuto.
Questo lavoro riporta i dati clinici relativi ad un paziente di sesso maschile di 8 anni di età giunto in pronto soccorso con quadro clinico di addome acuto e che ha necessitato di esplorazione chirurgica.
In Letteratura vengono segnalati pochi casi di addome acuto nel corso della MIS-C ma non sono disponibili ulteriori elementi sulla gestione del quadro clinico addominale e sulla sua evoluzione.
Gli autori rivisitano la propria esperienza alla luce dei dati di letteratura relativi alla MIS-C e riportano le evidenze cliniche che ne sono risultate.

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