

HUMAN CYTOMEGALOVIRUS INFECTION AND SUB-OCCLUSIVE INTESTINAL SYNDROME IN AN INFANT

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ABSTRACT

Human Cytomegalovirus is known to be the most common cause of congenital and perinatal infection in developed countries. The gastrointestinal involvement is not usually found among its' most frequent clinical presentations. We present the case of a male infant, aged 1 month and 2 weeks, admitted in our clinic presenting abdominal meteorism, with progressive intensification from the age of 3 weeks old, but normal passage of gas and feces. Laboratory tests revealed cholestasis syndrome, discrete hepatocytolysis syndrome and positive Ig M and Ig G levels for Cytomegalovirus. The imagistic investigations and surgical examination excluded the surgical origin of the symptoms. It was established the diagnosis of gastrointestinal pseudo-obstruction associated with perinatal Cytomegalovirus infection. During a second admission in our clinic, to exclude the possible complications of the Cytomegalovirus infection, a cranio-cerebral CT was performed which identified bilateral modification of otomastoiditis and raised the suspicion of moderate cerebral atrophy. Our case adds to the limited existing information on the clinical picture of Cytomegalovirus infection and supports the idea that the gastrointestinal manifestations of the infection are more common than known.

Key words: Human Cytomegalus infection, sub-occlusive intestinal syndrome, infant

INTRODUCTION

Human Cytomegalovirus (HCMV) is known to be the most common cause of congenital and perinatal infection in developed countries. The gastrointestinal involvement is not usually found among its' most frequent clinical presentations. Sub-occlusive intestinal syndrome is caused by a heterogenous group of enteric neuromuscular diseases that causes abnormalities of gut mobility. Sub-occlusive syndrome can occur at any age, but in young children, most often, can be fatal. This may be due to intrinsic primary or secondary visceral anomalies (drug toxicity, ischemia, inflammatory or autoimmune diseases, infection with Epstein Barr virus or Cytomegalovirus, myopathies). (1-3)

CASE REPORT

We present the case of a 1 month and 2 weeks old male (W= 4 500g, H= 56 cm, PC= 38,5 cm, IP= 0,90) transferred from another

hospital in the Pediatric Gastroenterology Clinic of „St Mary” Children’s Emergency Hospital for the appearance of abdominal meteorism, with progressive intensification from the age of 3 weeks, but normal passage of gas and feces. The toddler was born on term, with normal vaginal delivery, APGAR 9, birth weight 3150g, breast fed.

The clinical examination carried out at the time of presentation revealed impaired general status, jaundice of the skin and sclera, normotensive anterior fontanelle 2/1 cm, normal breathing and cardio beat rate, a distended, meteorised, firm abdomen, normal passage of gas, feces, semi consistent stool.

Laboratory tests revealed white blood cells number within normal limits, mild neutropenia associated to lymphocytosis and eosinophilia, hypochromic regenerative anemia, thrombocytosis, inflammatory syndrome absent, cholestasis syndrome (high levels of total bilirubin, with the initial predominance of the unconjugated fraction,

followed by the increase of the conjugated bilirubin, elevated alkaline phosphatase and GGT levels), discrete hepatocytolysis syndrome, amylase, glucozo-6-fosfat dehydrogenase, TSH, fT4, T4 within normal limits, LDH = 504 U/L, negative serology for hepatitis B, C, Epstein-Barr virus, and TORCH except for Cytomegalovirus, with positive Ig M and Ig G levels (HCMV IgG antibodies = 1.3915 UI/L, HCMV IgM antibodies positive). The urine examination was normal.

During the admission to our clinic the jaundice kept its intensity and the abdominal meteorism had a progressive intensification. Following the initial surgical assessment (Day 3) surgical intervention was not recommended.

The abdominal ultrasound identified intense air-dilated bowel loops.

The abdominal X-ray in dynamics revealed multiple loops of gas filling the bowel and a presacral air bubble (fig. 1). (3).

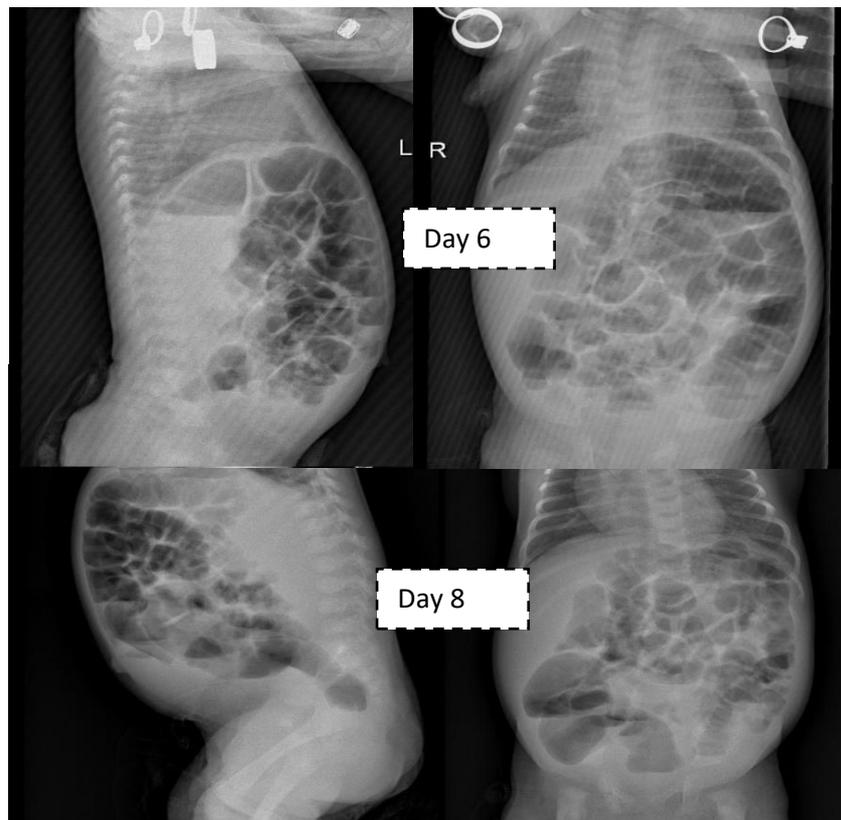


Figure 1. Abdominal X-ray in dynamics: multiple loops of gas filling the bowel, presacral air bubble

The surgical examination performed one week after the admission established the diagnosis of intestinal sub-occlusive syndrome (vomiting, intense meteorised abdomen, semi consistent stool). As a consequence, the toddler was transferred to Pediatric Surgery Clinic where during the admission (7 days) the patient had a slow favorable evolution - the abdomen gradually recovered his normal consistency and the

diagnosis of intestinal occlusion of surgical etiology was denied. The patient came back to our clinic for the management of the jaundice that had a slow remission.

With these findings, a diagnosis of gastrointestinal pseudo-obstruction associated with perinatal HCMV infection was established.

During the whole period the patient was breastfed and received hydro-electrolytic

rebalancing treatment, antibiotic therapy (Ceftriaxone), corticosteroids, Phenobarbital and for eliminating the intestinal gas was used a rectal catheter. Regarding the acute HCMV infection, since paraclinical evidence were within relatively normal limits, it led to the decision to follow the case without administering antiviral therapy.

With a stable evolution during admission to our clinic, the patient was discharged with the recommendations of breastfeeding, treatment with probiotics and Trimebutine for one month.

At the age of 3 months (W=6100g, T=61 cm, CP=40cm, IP=0.92) the toddler came back to our clinic for a clinical-biological reevaluation. On examination the general status was relatively good, without signs of jaundice or fever, mild pharyngeal congestion, generalized micro-

polyadenopathies, normotensive anterior fontanelle 2/1 cm, normal cardiopulmonary assessment, mild meteorized and painless abdomen, physiological intestinal transit, normal diuresis.

The blood tests revealed lymphocytosis, discrete hepatocytolysis syndrome, Cytomegalovirus with persisting high Ig G and Ig M levels.

The abdominal ultrasound identified massive aerocoly.

A neurological exam was performed and it recommended a cranio-cerebral CT to exclude the possible complications of the Cytomegalovirus infection. The cranio-cerebral CT showed bilateral modification of otomastoiditis and raised the suspicion of moderate cerebral atrophy (fig. 2), so a contrast-enhanced cranio-cerebral IRM follow-up over 6 months was recommended.



Figure 2. Cranio- cerebral CT- moderate cerebral atrophy

The patient was discharged with the recommendation of breastfeeding and reevaluation over 2 months.

DISCUSSION

In recent years the understanding of HCMV infection has improved due to the increased awareness of its consequences and to the improved diagnostic tests. The most common clinical presentations of the congenital or perinatal infection include microcephaly, intrauterine growth restriction, hepatosplenomegaly, jaundice, petechiae, thrombocytopenia or conjugated hyperbilirubinemia, intracerebral

calcification, chorioretinitis and evidence of hearing loss on newborn screen. The gastrointestinal involvement is not very frequent (1, 2).

We found several reports of cases of HCMV infection in neonates/toddlers associated with gastrointestinal presentations. The most frequent entity associated with HCMV infection was HCMV enterocolitis presented as necrotizing enterocolitis (NEC) with symptoms as diarrhea, fever and abdominal distension. There was evidence of pathologic condition related to Meckel's diverticulum (volvulus and perforation, respectively), ileal and colonic strictures and

obstruction. All cases had pathologic evidence of HCMV infection (e.g., bowel specimens with inclusion bodies and/or immunohistochemically stains positive for HCMV) or positive urine culture. (2)

There is also a hypothesis that HCMV-induced proinflammatory could cause the interruption of the gut's normal development or could in turn lead to a disruption in the normal development of the gut and malrotation as a result - a few cases of coexistent congenital Cytomegalovirus infection with intestinal malrotation and positive intestinal Cytomegalovirus biopsy were reported on several reviews. (4)

There is also proof of association between the congenital HCMV infection with Hirschsprung's disease. Aganglionosis was found in the resected bowel specimens of children whose serologic data, urinary viral culture and gene expression by in situ hybridization suggested a HCMV infection (5).

Our case adds to the limited information regarding the recognition of gastrointestinal symptoms in relation to HCMV infection. We

CONCLUSION

Our case adds to the clinical spectrum of HCMV infection and supports the idea that gastrointestinal presentations of the disease are more common than recognized.

presented a toddler with congenital HCMV infection confirmed by positive serology, whose clinical manifestation were jaundice and anemia unusual associated to an intestinal sub-occlusive syndrome spontaneous resolved and imagistic evidence of cerebral atrophy.

For the differential diagnosis of the sub-occlusive intestinal syndrome we considered the congenital causes (anorectal malformation, Hirschsprung's disease, Meckel's diverticulum, jejunoileal atresia, malrotation, infantile hypertrophic pyloric stenosis, meconium ileus, duodenal atresia, annular pancreas, chronic intestinal pseudo-obstruction idiopathic/secondary) and the acquired causes (intussusception, post-op adhesions, ileo-cecal tuberculosis, paralytic ileus, worm obstruction, obstructed inguinal hernia). (6)

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