



A case report of newborn with Klippel-Trenaunay syndrome from mother currently infected by COVID-19

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Background: Coronavirus disease 2019 (COVID-19) novel severe acute respiratory syndrome affected the world population with an infectious condition for which therapeutic forms are limited and vascular sequelae are major challenges. Pregnant women are physiologically immunocompromised and can be more affected by the pandemic than the general population. Extra precaution measures against COVID-19 during and after the outbreak are essential to ensure the safety of the newborn. The aim of the present study was to report the cesarean birth of a female child with Klippel-Trenaunay of a mother currently infected by COVID-19.

Case Description: A newborn of a 21-year-old mother with COVID-19 in the final stage of the disease was born through a cesarean section at full term. The physical examination at birth revealed substantial edema of the left lower limb associated with port-wine stains on the limb and left torso. The ultrasound revealed venous-lymphatic malformation affecting the entire left lower limb, suprapubic region and ipsilateral abdominal region. Two evaluations for COVID-19 were performed and both were negative. Infants of mothers with active COVID-19 infection during the birth period have a low probability of being positive for the disease, but precautions should be taken.

Conclusions: The association with malformations, such as the rare Klippel-Trenaunay syndrome (KTS), constitutes another challenge regarding the diagnosis and proper conduct to be taken with these children.

Keywords: Newborn; Klippel-Trenaunay syndrome (KTS); mother; coronavirus disease 2019 (COVID-19); case report

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Introduction

Coronavirus disease 2019 (COVID-19) novel severe acute respiratory syndrome affected the world population with an infectious condition for which therapeutic forms are limited and vascular sequelae are major challenges (1,2). Pregnant women are physiologically immunocompromised and can be more affected by the pandemic than the general population. Extra precaution measures against COVID-19 during and after the outbreak are essential to ensure the safety of the newborn and healthcare providers (3). Infants at risk should be transported in an incubator and tested for COVID-19 in a negative pressure room immediately after the bath. Skin-to-skin contact, and breastfeeding should continue under certain circumstances (4,5).

In a sample of 42 newborns of mothers infected by COVID-19, the polymerase chain reaction (PCR) was positive in one case and negative in the rest. Thus, vertical transmission remains inconclusive (6). A meta-analysis involving pregnant women with confirmed COVID-19 detected a 1.6% rate of stillbirths and neonatal deaths, whereas 3.5% of livebirths were positive for COVID-19 (7). Another meta-analysis found a 2.94% greater risk of the incidence of infection by severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) in non-separated newborns compared to separated newborns, which is not a statistically significant difference (8).

Klippel-Trenaunay syndrome (KTS) is a congenital

vascular disease characterized by cutaneous hemangiomas, varicose veins, and hypertrophied limbs (1). The estimated incidence is two to five per 100,000, with a greater frequency in males than females and no racial predilection (9). Extra-trunk lymphatic malformations are found in 21% of patients and trunk lymphatic malformation are found in 28% (10).

The aim of the present study was to report the cesarean birth of a female child with Klippel-Trenaunay of a mother currently infected by COVID-19. We present this case in accordance with the CARE reporting checklist (available at <https://acr.amegroups.com/article/view/10.21037/acr-23-193/rc>).

Case presentation

All procedures performed in this study were in accordance with the ethical standards of the Ethical Committee of Medicine School of Sao Jose do Rio Preto-Brazil (No. 4.987.427) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the mother patient for publication of this case report.

A newborn of a 21-year-old mother with COVID-19 in the final stage of the disease was born through a cesarean section at full term. Somatic status: 38 weeks, Apgar 9/10, weight 3,345 g, cephalic perimeter 30 cm, stature 50 cm. The infant presented strong crying and good flexion tonus, with no need for airway aspiration or other interventions. The physical examination at birth revealed substantial edema of the left lower limb associated with port-wine stains on the limb and left torso. The ultrasound revealed venous-lymphatic malformation affecting the entire left lower limb, suprapubic region and ipsilateral abdominal region. Two evaluations for COVID-19 were performed and both were negative.

Discussion

The present study reports vascular malformation (Klippel-Trenaunay) in a newborn girl from a mother currently infected by COVID-19. The two exams of the child for COVID-19 were negative and the finding of KTS was what most stood out during the physical examination. The prevalence of positivity for COVID-19 cited in the literature is low among newborns, but care regarding possible complications should be taken during the birth procedure (5,6). The literature suggests maintaining the infant with the mother, but preventive care should be employed (4).

Although newborns infected by COVID-19 often have

Highlight box

Key findings

- In this report, we report a newborn with Klippel-Trenaunay of a mother currently infected by coronavirus disease 2019 (COVID-19).

What is known and what is new?

- Some published studies confirm that the course of COVID-19 in pregnant women resembles that of other populations. However, there is no study associated vascular malformation (Klippel-Trenaunay).
- This is an association that may provide further evidence for new study about pathophysiological mechanisms with maternal viral infection by COVID-19 in relation with vascular malformations.

What is the implication, and what should change now?

- Important alert even if the child is negative for COVID-19 and the syndrome is not related, it is necessary to maintain preventive care in the first year together with the mother to avoid the risk of complications and death.

symptoms that mimic sepsis and one-third of affected patients may require some form of respiratory support, the short-term prognosis is favorable and the majority recover within 2 weeks after the onset of symptoms (4). In the present study, the infant was negative for COVID-19, but the occurrence of KTS raises a question regarding whether there is an association. KTS is a congenital malformation with the possibility of an associated clinical condition of lymphedema, indicating a deficiency of the lymphatic system in part of the body. The observance of edema during the physical examination may be a component of limb hypertrophy, which is a common finding in the syndrome, and suggests the presence of lymphedema since birth.

In observational clinical practice regarding patients with lymphedema that contracted COVID-19, many reported experiencing pain and an increase in the edema of the affected limb. This information serves as an alert for this type of complication for children born with COVID-19 associated with KTS. The conduct regarding this syndrome is initially watchful waiting. The complications that emerge should be assessed and treated when indicated.

Conclusions

Infants of mothers with active COVID-19 infection during the birth period have a low probability of being positive for the disease, but precautions should be taken. The association with malformations, such as the rare KTS, constitutes another challenge regarding the diagnosis and proper conduct to be taken with these children.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at <https://acr.amegroups.com/article/view/10.21037/acr-23-193/rc>

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <https://acr.amegroups.com/article/view/10.21037/acr-23-193/coif>). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the Ethical Committee of Medicine School of Sao Jose do Rio Preto-Brazil (No. 4.987.427) and with the Helsinki Declaration (as revised 2013). Written informed consent was obtained from the mother patient for publication of this case report. A copy of the written consent is available for review by the editorial office of this journal.

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