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CASE REPORT PEER REVIEWED | OPEN ACCESS

Complete thrombosis of the abdominal aorta in a neonate with advanced lower limbs ischemia: Etiological diagnosis, prognosis, and surgical approach

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ABSTRACT

Introduction: Neonatal aortic thrombosis is a rare but some cases have been described in the literature. It is associated with high mortality. The main etiology is umbilical vessel catheterization, but others cause coagulopathy, so sepsis could not be excluded. We reported a case of thrombosis of the abdominal aorta at the advanced stage of bilateral lower limbs ischemia treated surgically.

Case Report: At 41-week gestation, a newborn boy was born by vaginal delivery to a 28-year-old gravida II para II mother. He was transferred to the neonatal intensive care unit for neonatal icterus; fever associated with lower limbs' (extremities') coldness. Perinatal history was normal. Examination showed cold, cyanotic lower left limb, the right side was a cold, pale with toes necrosis. Bilateral abolition of femoral artery's pulses, loss of sensibility was found. At the left limb there was mild to moderate motor impairment. No history of umbilical artery catheterization was reported. A contrast computed tomography (CT) scan revealed complete aortoiliac thrombosis. Echocardiogram revealed normal anatomy without intracardiac thrombus. Preoperative blood analysis showed hemostasis anomalies. Cytobacterioligical urine exam was found to be positive for *Escherichia coli*. The baby underwent aortoiliac thrombectomy via median laparotomy. A surgical procedure was successful with blood flow restoration and limb perfusion. Post-operative blood count showed signs of ischemia-reperfusion, which may cause an unfavorable post-operative survival in our baby.

Conclusion: In conclusion, thrombosis of the aorta in neonates is rare with multiples etiologies. Surgical thrombectomy can be performed uneventfully with the best recanalization of the aorta. Early diagnosis and adequate post-operative management are the determinants of surgical outcomes.

Keywords: Abdominal aorta, Advanced limb ischemia, Ischemia-reperfusion, Neonatal thrombosis, Surgical thrombectomy

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INTRODUCTION

Aortic thrombosis in the neonate is a rare entity of uncertain etiology with a high mortality [1]. It is usually related to umbilical artery catheterization. The location is more commonly abdominal than thoracic [2]. We report a case of thrombosis of the abdominal aorta in a newborn treated at the advanced stage of lower limbs ischemia with a surgical approach.

CASE REPORT

At 41-week gestation, a newborn boy was born to a 28-year-old gravida II para II mother with two living children. The baby was born by vaginal delivery with neonatal parameters like Apgar scores of 9 and 10 at 1 and 5 min, respectively; birth weight was 3300 g, the other parameters were normal. Prenatal history was normal until the day of delivery. On day two after birth the baby was transferred to the neonatal intensive care unit for neonatal icterus, fever associated with coldness of lower limbs. Five days later, the evolution was marked by the intensification coldness of lower limbs (extremities), an apparition of cyanosis, pale, which extended bilaterally to limbs root. There was no history of umbilical vessels venous catheterization, no history of peripheral arterial catheterization.

Examination showed cold, cyanotic lower left limb; the right side was a cold, pale with toes necrosis. Bilateral abolition of femoral artery's pulses, loss of sensibility, delayed capillary refill in both limbs, and mild to moderate motor impairment to the left limb were found (Figure 1). There was no alteration of the conscious state, no heart murmur, heart rate was 135 beats/min, and respiratory rate was 48/min, temperature 38.2°C. Electrocardiogram found regular sinus rhythm. From that moment, heparin therapy was started.

As investigations, Doppler ultrasound showed an absence of flow in the abdominal tract of the aorta. A contrast CT scan of the thoracic, abdomen, and lower extremities revealed complete subrenal aortic thrombosis extending to both iliac arteries (Figure 2). An echocardiogram revealed normal anatomy without intracardiac thrombus. Chest radiography was normal.

Blood analysis revealed: platelet count was 38,000/mm^3, hemoglobin 21.2 g/dL, hematocrit was 69.2%, white blood cells at 12,250 mm^3, urea 0.82 g/L, creatinine 3.1 mg/L, sodium 141 mmol/L, kaliemia 5.2 mmol/L. The sedimentation velocity was 2 at the first hour and 5 at the second hour. C-reactive protein came at 10 mg/L, prothrombin time (PT) count 60.8% with an INR of 1.2, fibrinogen came at 2.43 g/L. Liver exploration found total bilirubin 236.36 mg/L, direct bilirubin 223.13 mg/L, conjugated bilirubin 13.13 mg/L, aspartate transaminase (ASAT) 219 IU/L, alanine transaminase (ALAT) 72 IU/L. Cytobacterioligical urine exam was found to be positive of *Escherichia coli*. Antibiotic had been administrated according to the isolated germ.

Surgical approach

Urgent surgery was decided after a surgical team discussion about vital and functional prognosis. The baby was induced under general anesthesia and monitoring lines are placed. End tracheal tube is placed. Through a median laparotomy, the aorta and both iliac arteries were controlled. A single transverse aortotomy just above its bifurcation was performed. We started with a thrombectomy of the aorta, then with 3 Fr balloon catheter, thrombectomy of both iliac arteries and the femoro-popliteal axes of both limbs were performed (Figure 3). There was no evident intestinal necrosis. Restoration of flow through the aorta was obtained after thrombectomy. The aorta was closed. Post-operative examination showed presence of left femoral pulse. The right femoral pulse was not found. The skin of the right lower extremity (limb) was found to be perfused (Figure 4). Therapeutic unfractionated heparin was continued post-operatively with continuous infusion.

Immediate post-operative blood count showed: Blood count revealed: platelet count was 30,000/mm^3, hemoglobin 20. 9 g/dL, hematocrit was 68.30%, white blood cells 13,580 mm^3, urea 0.49 g/L, creatinine 3.8 mg/L, sodium 138 mmol/L, kaliemia 6.5 mmol/L, bicarbonate 10 meq/L. Liver exploration found total bilirubin 236.36 mg/L, ASAT 244 IU/L, ALAT 58 IU/L. The diuresis was conserved. The hyperkalemia and acidosis had been corrected.

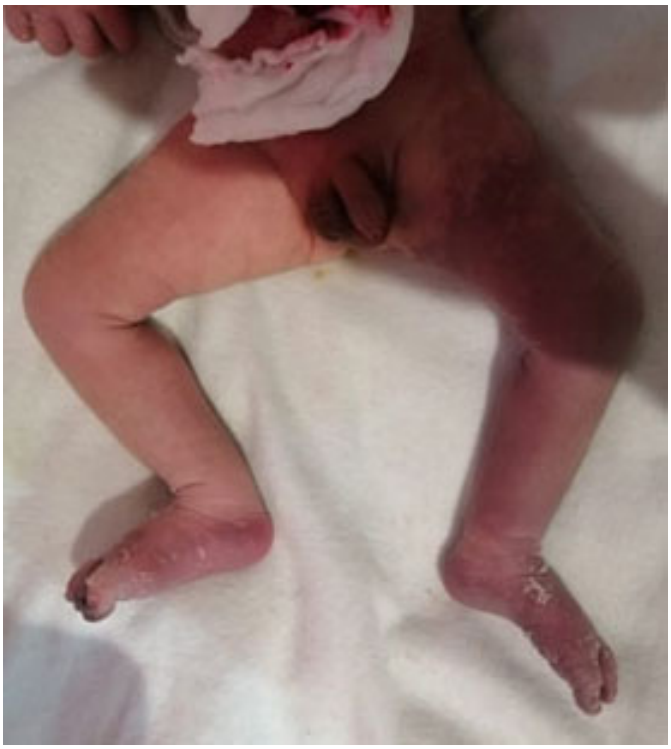

Figure 1: Showing bilateral lower limbs ischemia.

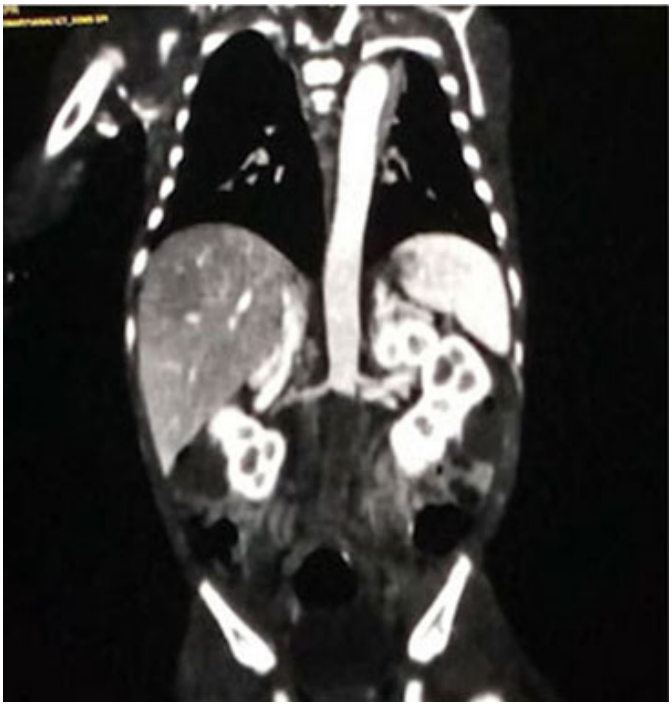

Figure 2: CT scan showing abdominal aorta thrombosis.

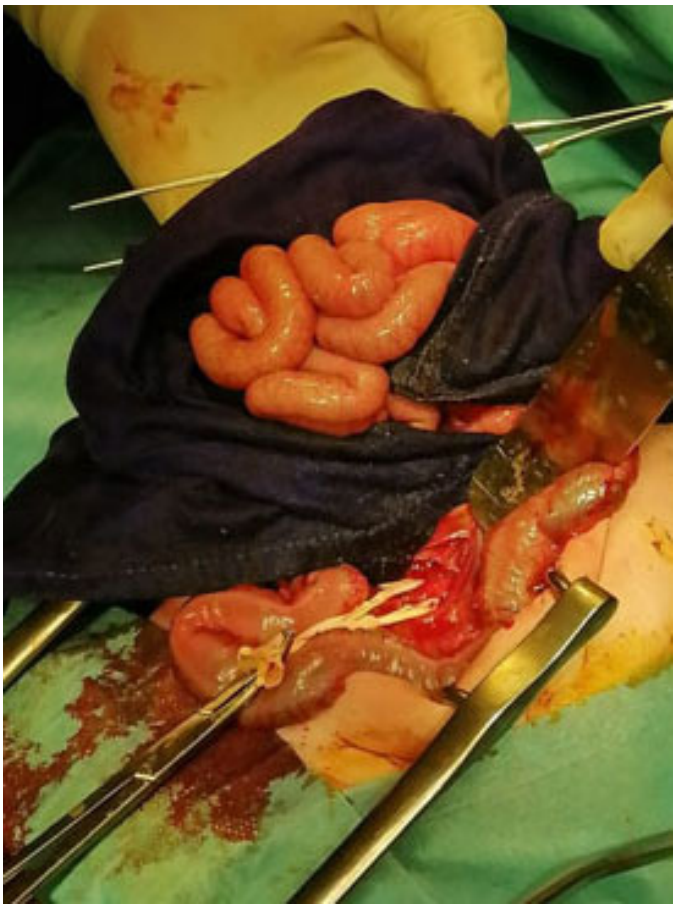

Figure 3: Intra-operative view of the abdominal aorta.

At 24 hours, post-operative blood exploration revealed urea at 0.49 g/L, creatinine 3.8 mg/L, sodium 138 mmol/L, kaliemia 9.92 mmol/L, and alkaline reserve at 7 meq/L, PT came at 12%; the activated thromboplastin time was prolonged at 100 second, hemoglobin 9.7 g/dL after exsanguinations. The correction of these anomalies was initiated. We could not organize renal hemodialysis. The suites had been marked by cardio respiratory arrest. Cardio-respiratory resuscitation was initiated but the baby was diseased one-hour following.

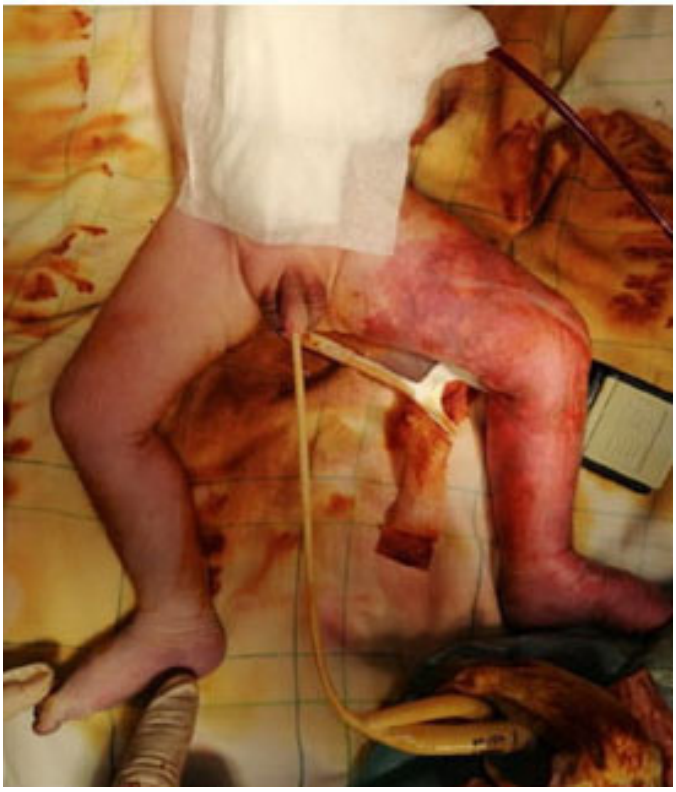

Figure 4: Post-operative picture showing limbs reperfusion.

DISCUSSION

Predisposing factors to aortic thrombosis include umbilical artery catheterization [3], maternal diabetes [4], polycythemia, hypernatremic dehydration [5], asphyxia, and patent ductus arteriosus [2], coagulopathy [3, 6]. Umbilical artery catheterization is the most common predisposing factor. A indwelling umbilical catheter acts as the single greatest risk factor of aortic thrombosis, and several additional factors such as dehydration and septicemia increase the risk of thrombosis [7, 8]. Other studies reported that the venous and arterial systems are equally involved in symptomatic neonatal thrombosis [8].

Otherwise, an inherited thrombophilia plays a role in thrombus formation in neonates. Some studies include congenital thrombophilia as risks factors. Wieland et al. [9] found factor V Leiden mutation or protein C deficiency as risk factors in the development of aortic thrombosis in neonate. According to Piersigilli et al. [10], an inherited thrombophilia, genetic factors, such as

the factor V Leiden, antithrombin, protein C or protein S deficiency, the 20210A prothrombin mutation could not be excluded in the pathogenesis of this pathology. Additionally, certain cases due to sepsis in which the isolated germ was *Klebsiella pneumoniae* were reported by Kamate et al. [11].

In our case, there was not a history of umbilical catheterization. Infection at *E. coli* and polycythemia was found in the investigations. This shows that the thrombosis is more likely due of sepsis coupled with coagulopathy. We did not perform C and S protein analysis because heparin therapy in the emergency department was started before any blood investigation.

Treatment options consist in anticoagulation with unfractionated heparin or low molecular weight heparin, systemic thrombolytic therapy and surgical thrombectomy. The treatment for neonatal thrombosis is still controversial, no guidelines currently exist [12]. However, the efficacy of thrombolytic therapy has been demonstrated. Monagle et al. [12] reported two cases treated successfully with low molecular weight heparin (LMWH). Other authors have reported improved outcomes with medical options [11–13].

Surgical thrombectomy also remains a therapeutic option with good results in certain cases [14, 15]. Our case was specific because of the advanced stage of the ischemia (Rutherford II-b on the right limb, Rutherford III one the left limb) with toes necrosis in contrast with the cases reported by Dieffnbach et al. [14] and Lofland et al. [15], respectively. Two reasons explained our decision despite the advanced stage. Our option was to save the right limb and avoid major amputation on the other side. The surgical procedure was successful in our case with recanalization of the aorta and iliac axes. The death was more likely due to the ischemia-reperfusion, according to post-operative blood analysis. Our treatment option is consistent with the literature. In fact, for patients with limb-threatening ischemia evidenced by gangrene, tissue loss or paralysis, recommendations are for therapeutic anticoagulation and upfront tissue-type plasminogen activators (tPA) thrombolysis followed by surgical thrombectomy if thrombolysis is unsuccessful. Within this construct, upfront surgery is only recommended for patients with contraindications to tPA [2]. Dieffnbach et al. [14] recommend therapeutic anticoagulation with or without systemic thrombolytic in cases with clinical evidence of ischemia still without imminent tissue loss or visceral injury. We conclude that clinicians should balance the risks and benefit of their decision to treat with the level of local expertise.

CONCLUSION

In conclusion, thrombosis of the aorta in neonates is a rare with multiples etiologies. Surgical thrombectomy can be performed uneventfully with the best recanalization of the aorta but best outcomes are associated with early diagnosis and adequate postoperative management.

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Author Contributions

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Issaka Zallé – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be 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

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.