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J Ped Surg Case Reports 4 (2016) 42–45

Contents lists available at ScienceDirect

Journal of Pediatric Surgery CASE REPORTS

journal homepage: www.jpscasereports.com

Aortic thrombus causing ischemic bowel mimicking as necrotizing enterocolitis in a premature neonate: A case report

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ARTICLE INFO

Article history:
Received 13 August 2015
Received in revised form
20 November 2015
Accepted 26 November 2015

Key words:
Aortic thrombus
Ischemic bowel
Enoxaparin

ABSTRACT

Spontaneous neonatal aortic thrombus is rare. Clinical presentation can vary depending on the location of the thrombus. We report a case of a premature infant with ischemic bowel likely due to an aortic thromboembolic event. The infant had presented with an acute abdomen, and underwent an exploratory laparotomy for concerns of worsening necrotizing enterocolitis. Intra-operative findings were suggestive of segmental ischemic small bowel with thrombosis of mesenteric vessels and a subsequent Doppler ultrasound revealed an infra-renal aortic thrombus. He was treated with subcutaneous enoxaparin, with effective reduction in thrombus size on serial scans. However, this treatment was complicated by bilateral subdural hematoma, and hence enoxaparin was discontinued. Aortic thrombus, although rare, should be considered in the differential diagnosis of an acute abdomen. Management of neonatal aortic thrombus remains controversial and further studies are required to aid clinicians in deciding the best management plan with minimal risk and optimal outcome.

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In a premature neonate presenting with abdominal distension and abdominal wall erythema, the most likely differential diagnosis would naturally be necrotizing enterocolitis (NEC). However, other less common causes such as aortic thrombus should also be considered. We present a case with a similar presentation attributed to bowel ischemia likely secondary to an infra-renal aortic thrombus.

1. Case report

This is a premature neonate with gestational age of 25 + 6 weeks who was born to a 27-year-old Gravida 1 Para 0 mother. Her antenatal history was complicated by cervical incompetence at 23 + 1 weeks and she had undergone cervical cerclage. The neonate was delivered prematurely in view of maternal chorioamnionitis as well as preterm labor.

His post-natal course was complicated by hyaline membrane disease requiring intubation and surfactant. He also had a large patent ductus arteriosus (PDA) complicated by pulmonary hemorrhage that failed 2 courses of Indomethacin and eventually required PDA ligation. He was then extubated to nasal intermittent mandatory ventilation (NIMV) on day 37 of life, and converted to continuous positive airway pressure (CPAP) on day 50 of life. In his immediate post-natal period, an umbilical arterial catheter (UAC) was inserted and it was removed after 7 days.

On day 72 of life, the infant was noted to have marked respiratory distress associated with desaturations and abdominal distension. He was kept nil by mouth (NBM), intubated and even required escalation to high frequency oscillatory ventilation (HFOV). Blood investigations revealed a high C-reactive protein (CRP) with neutropenia and thrombocytopenia requiring platelet transfusions. He was started on intravenous (IV) Cloxacillin, Amikacin and Metronidazole, and subsequently expanded to broader coverage with Meropenem, IV Gentamicin and IV Vancomycin.

The infant was also referred to the Pediatric surgeons in view of possible necrotizing enterocolitis (NEC) versus septic ileus. Serial abdominal X-rays (AXR) showed distended bowel loops but no obvious intramural gas (Fig. 1). However, the abdominal distension worsened and was associated with peri-umbilical and flank erythema with absent bowel sounds. He subsequently became hypotensive, requiring fluid boluses and inotropic support with dopamine infusion. An abdominal ultrasound showed free intraperitoneal fluid and inter-loop fluid with internal echoes and septations likely due to intra-abdominal infection. In view of

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http://dx.doi.org/10.1016/j.epsc.2015.11.013

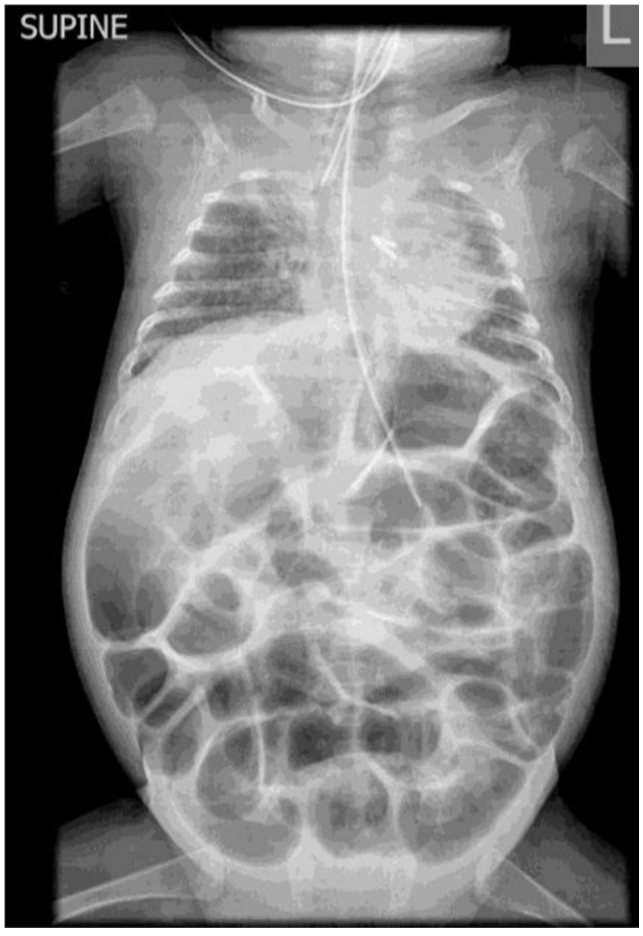

Fig. 1. AXR showing distended bowel loops.

worsening clinical status with increasing abdominal distension and abdominal wall erythema, along with an increasing trend of CRP from 120 to 265, the infant underwent exploratory laparotomy on day 74 of life.

The intra-operative findings revealed 48 cm of dilated, ischemic, non-viable small bowel which was resected. The segment was thin-walled with no visible peristalsis, and no perforations were noted. The supplying mesenteric vessel was found to be thrombosed, resulting in a wedge-shaped pattern of mesenteric infarction (Figs. 2–4). An ileostomy was created, and the infant had a remainder of healthy proximal 50 cm of small bowel from the duodeno-jejunal junction as well as 18 cm of terminal ileum distal to the resected margin, with preservation of the ileo-cecal valve. Post-operatively, the infant was kept nil by mouth (NBM) for 10 days before feeds were restarted and increased gradually as tolerated. However, his feeding was complicated by persistently high stoma output once he reached 100 mls/kg/day, and distal refeeding was initiated.

In view of the interesting finding of a thrombosed mesentery along with ischemic gut with no evidence of NEC, further investigations were conducted to search for evidence of intra-abdominal thrombi. A Doppler abdominal ultrasound showed a 1.1 cm long echogenic focus in the lumen of the infra-renal aorta which is suspicious for thrombus (Fig. 5). A repeat ultrasound one week after showed that the thrombus had grown to 2.2 cm. As such, decision was made to initiate treatment with subcutaneous enoxaparin. Prior to starting Enoxaparin, blood tests and a cranial ultrasound were done to ensure normal platelet levels and coagulation profile as well as absence of any intracranial hemorrhage. Of note, a 2D echocardiogram done on the day prior to the laparotomy did not demonstrate any evidence of right atrial thrombous or coarctation of the aorta (Fig. 5).

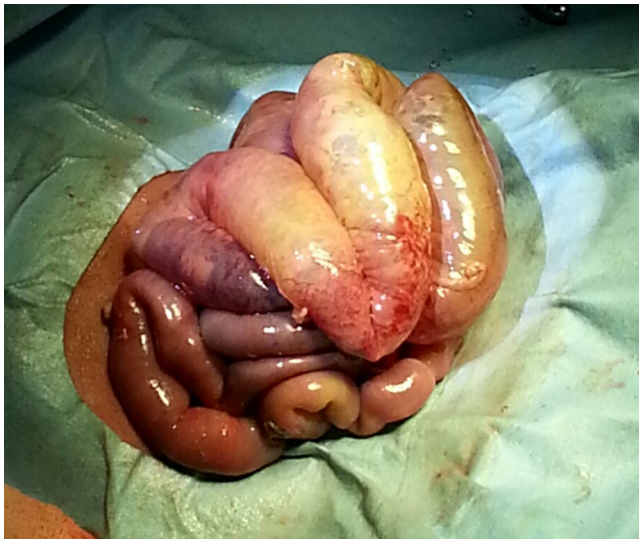

Fig. 2. Intra-operative finding of dilated proximal bowel and ischemic bowel segment.

It took 5 days to achieve therapeutic levels of anti-Factor Xa within 0.5–1.0 IU/ml, requiring a dose of 4.6 mg/kg/day of Enoxaparin given 12 hourly. Once treatment with Enoxaparin was initiated, the size of the thrombus remained stable. Anti-Factor Xa levels and platelet levels were monitored weekly. However, a cranial ultrasound done one month after starting treatment with Enoxaparin showed a left subdural hematoma measuring

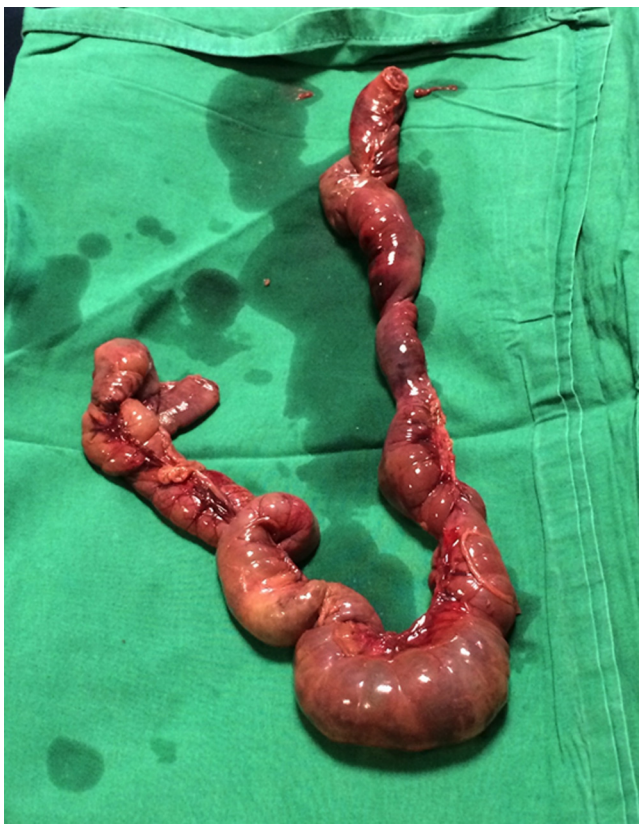

Fig. 3. Resected non-viable ischemic bowel segment.

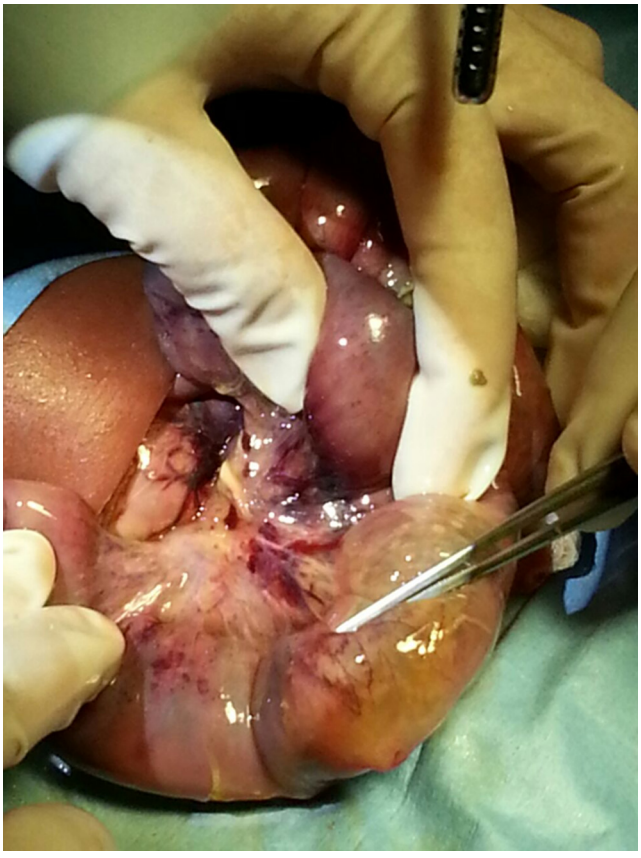

Fig. 4. Intra-operative finding of thrombosed mesentery.

0.4 × 2.8 × 1.2 cm despite having anti-Factor Xa levels within the therapeutic range as well as normal platelet levels. Given such complication, enoxaparin was discontinued. Weekly serial Doppler abdominal scans showed gradual reduction of the size of the thrombus, with the latest size being 1.3 cm. A follow-up cranial ultrasound three weeks later demonstrated a small right subdural hematoma as well, with the previously noted left subdural hematoma being stable in size. There were no other bleeding manifestations noted.

The infant underwent stoma closure on day 142 of life and is currently stable. A repeat Doppler ultrasound done at corrected age of about 9 months showed a fairly stable non-occluding echogenic thrombus measuring 0.2 cm. The latest Doppler ultrasound done at corrected age of 1 year old showed no further evidence of the aortic thrombus.

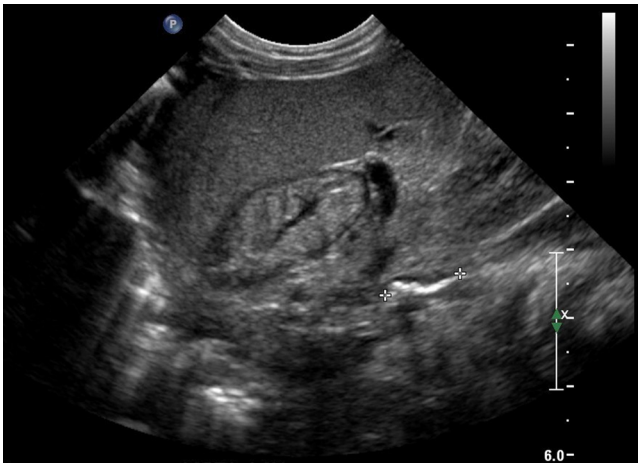

Fig. 5. Doppler ultrasound showing echogenic focus in the aorta suspicious for aortic thrombus.

2. Discussion

Neonatal arterial thrombosis is rare, and the incidence of symptomatic neonatal arterial thromboembolic disease has been reported to be 0.25 out of 10,000 live births [1]. Neonates tend to be in a physiological 'relative' pro-thrombotic state due to their immature hemostatic systems, thus increasing their susceptibility to thrombosis [2].

In the above case, given the surgical operative findings as well as ultrasound results, we postulate that the small bowel ischemia is secondary to the aortic thrombus, which had initially been causing mesenteric ischemia and may have subsequently migrated during or after the surgery to the infra-renal region of the aorta.

In neonates, up to 90% of arterial thrombosis are associated with arterial access devices [1]. A prospective study found that arterial thromboses were found in 23.4% of 47 babies who had a UAC inserted, although majority of these infants were asymptomatic [2]. The pathophysiology of thrombosis is related to vascular obstruction, endothelial damage by the catheter and low blood flow [3]. Spontaneous arterial thromboses are extremely rare and tend to be severe and mostly involve the aorta [2].

Apart from arterial access catheters, critical illness is also a well-recognized risk factor for thromboembolism due to the development of a pro-thrombotic state as coagulation inhibitors are being consumed [4]. Several other factors predisposing a neonate to thrombosis include congenital thrombophilia, congenital heart disease, dehydration, polycythemia and surgery [2,4].

In our infant, the cause of the aortic thrombus is unclear, but he did have risk factors that may possibly have contributed to formation of the thrombus, including UAC insertion, presumed sepsis and PDA ligation. His UAC was removed on day 7 and the clinical episode occurred on day 72 of life. Even though it is difficult to conclude whether the UAC could have been the source of the thrombus, a slowly evolving thrombus with subsequent embolization cannot be ruled out. Sepsis could be a potential cause, but blood cultures returned negative.

The clinical presentation of arterial thrombosis depends on the location of the thrombus, and include:

- Signs of limb ischemia (e.g. Pale or cold extremities, weak or absent pulses) especially related to cases of arterial catheterization of peripheral or central arteries
- Signs similar to that of necrotizing enterocolitis (e.g. Feed intolerance, bilious gastric aspirates, blood-stained stools) suggesting mesenteric infarction
- Increased blood pressure suggesting renal artery thrombosis
- Signs suggestive of arterial ischemia stroke (e.g. Seizures, lethargy) [4]

Management of an arterial thrombus still remains controversial, as published data in the literature is limited to small case series or case studies. A Cochrane review also did not provide any conclusions as no eligible studies were found [5]. The management includes clinical observation, anticoagulation, thrombolytic therapy or surgical thrombectomy. If the patient were asymptomatic, clinical observation may be indicated, but close regular monitoring is required and anticoagulation initiated if the clot were to enlarge [6].

With regards to anticoagulation, low molecular weight heparin (LMWH) is increasingly being used as compared to unfractionated heparin as it allows for subcutaneous application and has a lower

incidence of bleeding [6]. Preterm infants tend to require a longer time for anti-Factor Xa levels to reach target range as compared to term infants (6 versus 2 days) and also tend to require higher doses of enoxaparin (2.1 mg/kg/dose 12 hourly versus 1.7 mg/kg/dose 12 hourly) [4], as seen with our case. This might be due to neonates having a reduced response to heparin due to their low levels of antithrombin and a higher rate of heparin clearance [2].

Thrombolysis should only be considered in cases of limb or organ-threatening thrombosis and acute atrial clots [2]. In the neonatal population, recombinant tissue plasminogen activator (rt-TPA) is the most widely used agent as it has several advantages including a short half-life, minimal antigenicity, high fibrin specificity with poor activation of free plasmin [4]. Very close monitoring is also vital during the therapy and rt-TPA should be discontinued once the clot has lysed [2]. Surgical intervention should be the last option, with the main aim being to maximize the chances of limb salvage in the rare cases of life or limb-threatening thrombosis [4].

Nevertheless, all interventions are not without their risks and complications. The main concern with anti-thrombotic therapy in this population would be intracranial hemorrhage [2]. Therefore, prior to initiating treatment, the clinician should assess the infant and only proceed if the potential benefits outweigh the risks.

With regards to long-term outcomes and prognosis of neonatal aortic thrombosis, more information is still required, but Nagel et al. showed that 82% of neonates with aortic thrombosis survived till hospital discharge, and there was a mortality rate of 15% [7].

3. Conclusion

In conclusion, this article presents a case of neonatal aortic thrombus presenting with NEC-like symptoms, requiring treatment with enoxaparin. Therefore, in a neonate with such symptoms, aortic thrombus should also be considered as a differential diagnosis, especially when laboratory and radiographic findings are not strongly suggestive of NEC. In the neonatal population, management of neonatal arterial thrombosis remains controversial and further studies are warranted to guide clinicians.

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