

CASE REPORT

Successful pregnancies after neonatal aortic thrombosis, childhood arterial repair, and a deficient pelvic vasculature

Thomas Troëng^{1,2} , Agneta Bergqvist³ & David Bergqvist²

¹Blekinge Hospital, Karlskrona, Sweden

²Department of Surgical Sciences, Vascular Surgery, Uppsala University, Uppsala, Sweden

³Karolinska Institutet, Stockholm, Sweden

Correspondence

Thomas Troëng, Blekinge Hospital, 371 85 Karlskrona, Sweden. Tel: +46 733 268829; Fax: +4645520133; E-mail: thomas.troeng@ltblekinge.se

Funding Information

No sources of funding were declared for this study.

Received: 25 November 2017; Revised: 5 February 2018; Accepted: 28 February 2018

Clinical Case Reports 2018; 6(6): 1045–1047

doi: 10.1002/ccr3.1498

Key Clinical Message

Neonatal aortic bifurcation thrombosis can cause occlusion of iliac arteries causing abnormal pelvic vasculature and claudication in childhood. A bifurcation graft normalizes the perfusion of the legs but not of the pelvis. In a girl, this does not preclude successful pregnancies in adult life. It has not been reported before.

Keywords

Aortic bifurcation graft in childhood, childhood aortic surgery, neonatal aortic thrombosis, normal pregnancies in vascular abnormality, pelvic vascular abnormality and pregnancy.

Introduction

A 11-year-old girl had an aortic reconstruction for a neonatal aortic occlusion causing claudication. Postoperatively, the pelvis was perfused only by the median sacral artery and collaterals. At the age of 36, a normal pregnancy occurred. Delivery was uneventful by a cesarean section. Two more similar pregnancies.

Neonatal abdominal aortic thrombosis related to the use of umbilical artery catheters was first reported in 1968 [1]. Successful treatment with surgical thrombectomy was reported in individual cases and followed by similar reports of thrombolysis. There are no reports of the long-term outcomes of these procedures. Conservative treatment increases the risk for leg growth abnormalities and reno-vascular hypertension [2].

We inserted an aortic bifurcation graft in a 11-year-old girl with claudication due to an abdominal aortic occlusion, caused by an umbilical catheter-induced thrombosis in the neonatal period [3]. We can now report a 30-year follow-up of this procedure.

Case Report

This girl was born prematurely in 1974 with a weight of 1130 g and kept on a ventilator for 4 weeks. An umbilical artery catheter was used for blood sampling and nutrition. An ischemic left leg motivated removal of the catheter on day 25. No efforts were made for a more detailed diagnosis. Not until the age of eleven, she developed symptoms of claudication when walking uphill. An occlusion of the aortic bifurcation was diagnosed. Both legs were perfused through collaterals, most abundant on the right side, refilling the right external iliac artery and the left common femoral.

Thus, at this age, a side-to-end infrarenal Dacron 12 × 6 mm right external iliac-left common femoral graft was inserted followed by normalization of walking capacity and ankle pressures. No pharmacological treatment was given postoperatively.

The patient had metal staples inserted near the right knee joint later the same year to treat leg length discrepancy likely caused by insufficient perfusion.

Six years later, her growth had stretched the graft legs causing graft stenoses and decreased ankle–brachial indices but no subjective complaints. The Dacron graft was replaced with a PTFE 14×7 mm bifurcation graft anastomosed to the same distal native artery sites. A post-operative intravenous digital subtraction angiography showed the pelvic organs perfused only by an enlarged median sacral artery and collaterals (Fig. 1). The bed of the right internal iliac artery is fed by the right-sided branch of the median sacral artery and possibly also from the right deep circumflex iliac artery. On the left side, the collateral branch seems to connect to a less developed internal iliac bed. The ankle–brachial indices were 0.9 in both legs, and walking distance was unlimited [4].

Moreover, the patient had a congenital aplasia of the left kidney and repeated episodes of right flank pain related to a stenosis of the right ureter. This was successfully dilated the year after the aortic surgery. Recurrent stenosis of the ureter causing pain 17 years later motivated a laparoscopic renal pelvoplasty in 2009. The preoperative CT angiography demonstrated a patent aortic reconstruction (Fig. 2).

The patient had been told by her gynecologist that pregnancy was unlikely because of her pelvic vascular

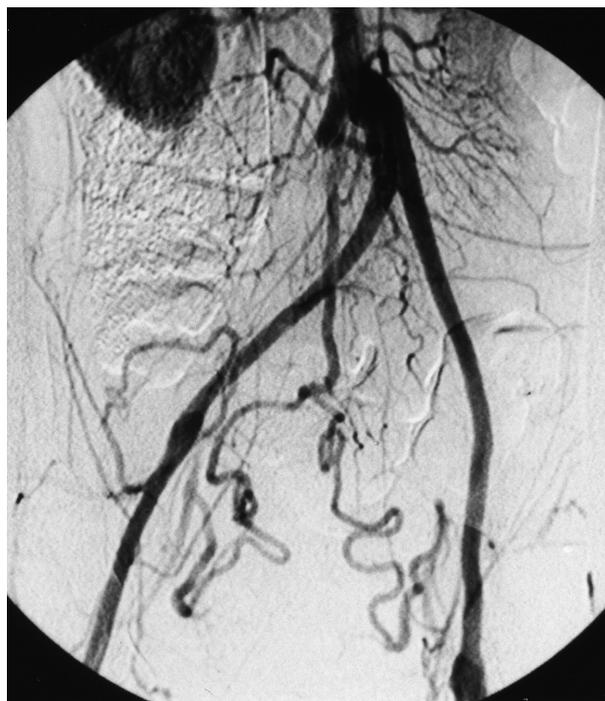


Figure 1. Intravenous digital subtraction angiography after graft replacement. Note the large median sacral artery and the continuing two collaterals. They appear to connect to the internal iliac systems, more so on the right side. The right side deep circumflex iliac artery may also contribute.

abnormality. Thus, when she became pregnant in 2010, it was unexpected. The pregnancy was uneventful with no symptoms of uterine ischemia. A healthy boy was delivered by cesarean section. At the operation, “wide vessels of the uterus” were noted. Two more healthy boys were born in the same way during the following 4 years. All three children were delivered in the 39th gestational week with normal birth weights, 3530, 4165, and 3610 grams.

At follow-up in 2016, the patient is a 42-year-old pre-school teacher with unlimited walking capacity and the mother of three healthy boys. Her ankle–brachial indices are 1.0 bilaterally. She has no medication and no planned follow-up.

Discussion

Interventional treatment of acute neonatal aortic thrombosis has been reported since the late 1960s. Early reports concerned open thrombectomy [5]; later thrombolysis was introduced also for this condition. Most publications



Figure 2. Abdominal CT angiography reconstruction at the age of 35, 19 years after the second aortic graft. The distal anastomoses are to the same native artery sites as the first graft, the right external iliac, and the left common femoral.

are case reports, and evidence for the choice of either method is lacking.

Our patient did not become symptomatic until her schoolyears. Surgical reconstruction of the abdominal aorta in childhood is uncommon, and most reports deal with the rare cases of fibromuscular dysplasia or Takayasu's disease. Evidence for preferring either open or endovascular surgery for aortic occlusion in children is lacking. In women treated with uterine artery occlusion or embolization for symptomatic fibroids, a study found 15 pregnancies in 102 women [6]. A possibly more extensive reduction in pelvic blood flow may be the result of bilateral internal iliac artery ligation for postpartum hemorrhage. In a recent report, three of 13 women thus treated got pregnant again within a two-year period [7]. Our patient had a more advanced arterial abnormality with a median sacral artery as the only antegrade vessel perfusing the pelvic organs through collaterals to the internal iliac systems making a normal pregnancy possible, although initially unlikely. Cesarean section was chosen for the deliveries as possible marginal uterine perfusion might cause fetal asphyxia during contractions.

Conclusion

This report demonstrates that pelvic vascular abnormality after neonatal aortic thrombosis in a girl and bifurcation graft insertion in childhood does not preclude successful pregnancies.

Conflict of Interest

The authors declare no conflict of interests.

Authorship

TT and DB: responsible for the clinical care over the years and wrote the manuscript. AB: contributed with the obstetric understanding of the case.

References

1. Stark, C. R., D. C. Abramson, and N. V. Erkan. 1968. Intravascular coagulation and hyaline-membrane disease. *Lancet* 2:520.
2. Seibert, J., F. Northington, J. Miers, and B. Taylor. 1991. Aortic thrombosis after umbilical artery catheterization in neonates: prevalence of complications in long-term follow-up. *AJR Am. J. Roentgenol.* 156:567–569.
3. Bergqvist, D., S. E. Bergentz, G. Hermansson, H. O. Lundberg, and T. Troëng. 1987. Late ischaemic sequelae after umbilical artery catheterization. *Br. J. Surg.* 74:628–629.
4. Troëng, T., D. Bergqvist, and S. E. Bergentz. 1994. Failing aortic graft in adolescence – an effect of patient growth. *Eur. J. Vasc. Surg.* 8:648–650.
5. Martin, J. E., J. F. Moran, L. S. Cook, K. K. Goertz, and L. Mattioli. 1989. Neonatal aortic thrombosis complication umbilical artery catheterization: successful treatment with retroperitoneal aortic thrombectomy. *Surgery* 105:793–796.
6. Firouznia, K., H. Ghanaati, M. Sanaati, A. H. Jalali, and M. Shakiba. 2009. Pregnancy after uterine artery embolization for symptomatic fibroids: a series of 15 pregnancies. *AJR Am. J. Roentgenol.* 192:1588–1592.
7. Domingo, S., A. Perales-Puchalt, I. Soler, B. Marcos, G. Tamarit, and A. Pellicer. 2013. Clinical outcome, fertility and uterine artery Doppler Scans in women with obstetric bilateral internal iliac artery ligation or embolization. *J. Obstet. Gynaecol.* 33:701–704.