Case Report

Takayasu Disease in Twin Pregnancy: A Case Report†

Jittima Rujiwetpongstorn MD*,
Yuri Yanase MD*

† *17th World Congress on Controversies in Obstetrics, Gynecology and Infertility, Lisbon, Portugal*
* *Department of Obstetrics and Gynecology, College of Medicine, Rangsit University, Rajavithi Hospital, Bangkok, Thailand*

Takayasu arteritis is a rare chronic inflammatory vascular disease involving the aorta and its major branches. During pregnancy, the disease can be life-threatening. This report describes a successful management of twin pregnancy complicated with Takayasu disease, rarely described elsewhere. A 33-year-old pregnant woman had been diagnosed with Takayasu disease since the age of 15 during a typical history and investigation. The patient underwent abdominal aortic aneurysmectomy with graft and revascularization of renal artery with saphenous vein graft before pregnancy. This pregnancy was a monochorion-diamnion twin. She was closely followed up and taken care of by a multidisciplinary approach at the high-risk antenatal clinic. A single fetal demise was detected at 20 weeks and the live fetus was diagnosed with intrauterine growth restriction at 25 weeks. Cesarean delivery was performed at 30 weeks due to severe IUGR, abnormal umbilical artery Doppler, and maternal superimposed preeclampsia, giving birth to a healthy female baby weighing 960 gm. The case presented here implies that a successful outcome of twin pregnancy complicated with Takayasu disease is possible with multidisciplinary approach and extreme cautions.

Keywords: *Takayasu disease, Twin pregnancy, Preeclampsia*

J Med Assoc Thai 2013; 96 (11): 1508-11
Full text. e-Journal: *http://jmat.mat.or.th*

Takayasu's arteritis (TA) is an idiopathic, chronic inflammatory vascular disease involving primarily the aortic arch and its major branches[1-3]. It affects predominantly females at reproductive years, in almost 80% of cases, with mean age of diagnosis at 29 years[4,5]. The disease can develop complications such as vascular stenosis, hypertension, or multiple organ dysfunction[2]. Pregnancy is relatively common in women with TA due to reproductive age and gender preference. However, pregnancy can deteriorate the disease and the disease, even stable, can adversely affect the outcome of pregnancy such as intrauterine growth restriction, fetal death, or pre-eclampsia. Even if pregnancies complicated with TA have been reported several times, the disease associated with twin pregnancies has not been described elsewhere. The objective of this report was to describe a successful management of twin pregnancy complicated with Takayasu disease. This case report has been approved by a suitably constituted Ethics Committee of the institution.

Correspondence to:
Rujiwetpongstorn J, Department of Obstetrics and Gynecology, College of Medicine, Rangsit University, Rajavithi Hospital, Bangkok 10400, Thailand.
Phone: 081-885-0088
E-mail: jittima_r@hotmail.com

Case Report

A 33-year-old woman, gravida 3, with previous miscarriage and preterm birth, first attended antenatal care clinic at 13 weeks of gestation. She had been diagnosed with Takayasu's arteritis at 15 years of age, presenting with clinical symptoms of hypertension, including severe headache, visual disturbance, palpitation, and abdominal mass. The physical examination revealed high blood pressure of 190/150 mmHg and midline abdominal mass 5 cm in diameter. Computer tomography demonstrated abdominal aortic aneurysm and bilateral renal stenosis. She underwent abdominal aortic aneurysmectomy with graft (Dacron) and revascularization of renal artery with saphenous vein graft, followed by medication control with antihypertensive drugs and prednisolone. At 26 years of age, surgical correction by aneurysmectomy was performed again and lateral patch repair owing to abdominal mass that was a false aneurysm of the left common iliac artery. At 30 years of age, she had a planned pregnancy since the disease was under control and a whole body check-up was normal. Nevertheless, the first pregnancy ended in an uncomplicated miscarriage at 11 weeks of gestation. A year later, she became pregnant again while the disease was stable but required treatment with antihypertensive drugs and prednisolone. The second

pregnancy was complicated with severe preeclampsia, ending up with a cesarean section due to a non-reassuring fetal status at 31 weeks of gestation, giving birth to a healthy female baby.

At 33 years of age, she had a third pregnancy. At the first visit at 13 weeks of gestation, she had high blood pressure, 180/110 mmHg. The ultrasound examination revealed a monochorion-diamnion twin pregnancy. She was admitted to control her blood pressure and an ultrasound evaluation of the intra-abdominal great vessels. The ultrasound findings were suggestive of aneurysm with thickening thrombus at the origin of the right renal artery. Laboratory values showed a positive result of antinuclear antibody-positive, anticentromere protein titer of greater than 1,280, 24-hour urine protein of 634 gm, creatinine 0.3 mg/dL. After comprehensive counseling on the risks and prognosis, the couple decided to continue with the pregnancy.

Her Takayasu's arteritis was closely taken care of by the cardiologist team. During pregnancy, her blood pressure became more difficult to control, requiring multi-antihypertensive drugs, including Atenolol, Prasozin, Aldomet, Amlodipine and Lasix and was maintained between150/90-160/100 mmHg. She also started ASA at 17 weeks of gestation.

The fetal growth of both fetuses was followed-up by a series of ultrasounds. A single fetal demise was detected at 20 weeks and the live fetus was diagnosed with intrauterine growth restriction at 25 weeks. The fetal growth and well-being was closely monitored by weekly ultrasound and Doppler ultrasound. At 30 weeks of gestation, severe intrauterine growth restriction and absent end diastolic flow of the umbilical arteries were detected. The patient's blood pressure was approximately 160/110 mmHg, in spite of proper medications. Laboratory results had slightly changed. Creatinine was rising to 1.0 mg/dL. Uric acid had increased to 8.8 mg/dL. The patient was admitted and diagnosed with pregnancy-aggravated hypertension. Dexamethasone and magnesium sulfate was given. Doppler ultrasound was performed daily to surveillance the fetal status. Occasional reverse end diastolic flow of the umbilical arteries was detected 3 days after being admitted. Cesarean section with tubal resection was performed due to severe intrauterine growth restriction, abnormal umbilical artery Doppler, and previous cesarean section, delivering a female baby weighing 960 gm. with good APGAR scores. The baby was taken care by the neonatologist at the newborn intensive care unit for nearly 2 months and was discharged in a healthy condition. Postpartum maternal condition showed no complication and the disease was controlled as was during the pre-pregnancy period. The CTA was performed at 8 weeks postpartum period as shown in Fig. 1 and 2.

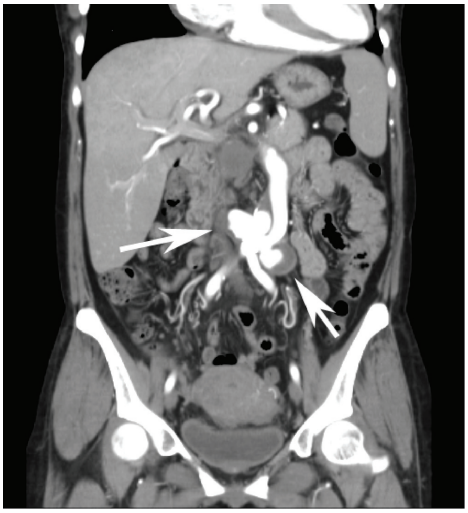

Fig. 1 Contrast enhanced CT of the whole aorta in coronal view show 2 lobulated saccular aneurysms at infrarenal abdominal aorta, just above aortic bifurcation (arrow). It is measured 3.5x2.9 cm in right side and 3.4x2.9 cm in left side. Partial thrombosis of aneurysm is present.

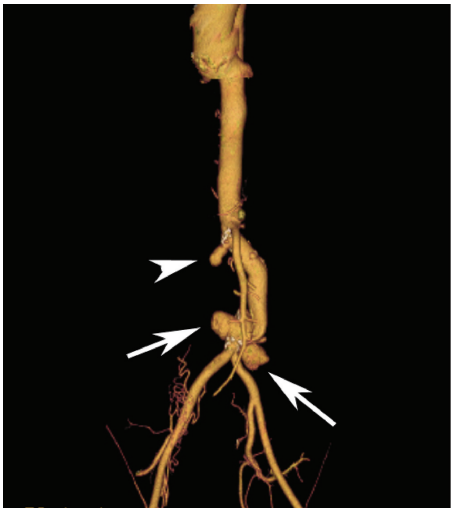

Fig. 2 Contrast enhanced CT of aorta with 3D reconstruction show saccular aneurysms at lower abdominal aorta, just above aortic bifurcation (arrow), and another saccular aneurysm in the right side of abdominal aorta at juxtarenal position (arrow head). There is severe stenosis of bilateral main renal arteries (not show in image).

Discussion

Takayasu's arteritis was firstly described in 1908 by a Japanese ophthalmologist, Mikito Takayasu. TA has worldwide distribution, with the highest incidence in Japan, India and Southeast Asia[6-9]. The cause is unknown, but it seems to be related to genetic factors as well as autoimmunity with the presence of histocompatibility antigens, particularly HLA A9, A10, B5, BW40, BW52, DQW1, DR2, DR4, DR7, DW3 and DW12[10]. The female to male ratio is 8:1[11]. Clinical manifestation of decreased pulsation and the differences of blood pressure between the arms, or bruits over the subclavian arteries or abdominal aorta may raise suspicion of the disease. Nevertheless, the diagnosis is usually based on CT, MRI, or angiography with typical findings of thickened vessel wall and tapered luminal narrowing or occlusion.

Because the incidence of Takayasu arteritis during childbearing years is relatively high, pregnancy is common among patients with TA. However, pregnancy complicated with the disease can be life-threatening and the management of which is challenging. Pregnancy with Takayasu arteritis has a higher risk of pregnancy-aggravated hypertension, as seen in the case presented here, complications due to the worsening of the cardiovascular. The increased intravascular volume due to physiological change during pregnancy can impair maternal hemodynamics and exacerbate the pre-existing hypertension, aortic regurgitation, cerebrovascular accident, and congestive heart failure[7,12,13]. The preexisting hypertension is often more severe especially in late pregnancy and often associated with intrauterine growth restriction, fetal hemorrhage, and maternal heart failure.

Although proper antenatal and intrapartum management with a multidisciplinary approach, the adverse pregnancy outcomes are still rather high[7]. However, a successful outcome could be expected if judicious medication and a multidisciplinary approach as well as a timely admission are instituted. The cornerstone of management is control of blood pressure, which plays an important role in pregnancy outcome.

Though several cases and a series of pregnancies with Takayasu arteritis have been reported, twin pregnancies complicated with Takayasu arteritis represent a clinical entity very rarely seen. To the best of our knowledge, this is the first case of twin pregnancy occurring in a woman with Takayasu arteritis. On a theoretical basis, twin pregnancy potentially carry an even higher risk of adverse outcomes, since twin itself could be associated with increase in blood volume and several complications such as fetal growth restriction or pregnancy-induced hypertension. Of note, the case presented here was associated with the demise of one twin the other twin with growth restriction and maternal pregnancy-aggravated hypertension. Therefore, this case signified that twin pregnancy with Takayasu arteritis is likely to be associated with even more adverse outcomes, though based on only one case. Nevertheless, the surviving twin in this case, in spite of preterm birth and growth restriction, suggests that a successful outcome is possible in twin pregnancy complicated with the disease, if the care of such patients is intensive and given by a multidisciplinary team, especially perinatologist taking care of mothers and fetuses and vascular surgeons providing input on cardiovascular implications both antepartum and postpartum periods.

Takayasu disease can cause many complications in pregnancy, to both mother and baby. Knowledge and understanding of the interaction between the two conditions allow for well-informed decision making and favorable outcomes of the pregnancy, as well as proper long-term follow-up and care with appropriate clinicians. A successful outcome of twin pregnancy complicated with Takayasu disease is possible with a multidisciplinary approach and extreme cautions.

What is already known on this topic?

Takayasu disease can increase the risk of adverse pregnancy outcomes as well as maternal complications.

What this study add?

A successful outcome in twin pregnancy complicated with Takayasu disease is possible to successful outcome with multidisciplinary approach and extreme cautions.

Potential conflicts of interest

None.

References

1. Hauenstein E, Frank H, Bauer JS, Schneider KT, Fischer T. Takayasu's arteritis in pregnancy: review of literature and discussion. J Perinat Med 2010; 38: 55-62.
2. Leal PC, Silveira FF, Sadatsune EJ, Clivatti J, Yamashita AM. Takayasus's arteritis in pregnancy. Case report and literature review. Rev Bras

Anestesiol 2011; 61: 479-85.
3. Railton A, Allen DG. Takayasu's arteritis in pregnancy. A report of 4 cases. S Afr Med J 1988; 73: 123.
4. Ioscovich A, Gislason R, Fadeev A, Grisaru-Granovsky S, Halpern S. Peripartum anesthetic management of patients with Takayasu's arteritis: case series and review. Int J Obstet Anesth 2008; 17: 358-64.
5. Kerr GS, Hallahan CW, Giordano J, Leavitt RY, Fauci AS, Rottem M, et al. Takayasu arteritis. Ann Intern Med 1994; 120: 919-29.
6. Vanoli M, Daina E, Salvarani C, Sabbadini MG, Rossi C, Bacchiani G, et al. Takayasu's arteritis: a study of 104 Italian patients. Arthritis Rheum 2005; 53: 100-7.
7. Mandal D, Mandal S, Dattaray C, Banerjee D, Ghosh P, Ghosh A, et al. Takayasu arteritis in pregnancy: an analysis from eastern India. Arch Gynecol Obstet 2012; 285: 567-71.
8. Hidaka N, Yamanaka Y, Fujita Y, Fukushima K, Wake N. Clinical manifestations of pregnancy in patients with Takayasu arteritis: experience from a single tertiary center. Arch Gynecol Obstet 2012; 285: 377-85.
9. Suri V, Aggarwal N, Keepanasseril A, Chopra S, Vijayvergiya R, Jain S. Pregnancy and Takayasu arteritis: a single centre experience from North India. J Obstet Gynaecol Res 2010; 36: 519-24.
10. Mwipatayi BP, Jeffery PC, Beningfield SJ, Matley PJ, Naidoo NG, Kalla AA, et al. Takayasu arteritis: clinical features and management: report of 272 cases. ANZ J Surg 2005; 75: 110-7.
11. Khalaf HH, Arafah MR, Refaat AA, Ibrahim MF. Coronary artery bypass grafting for Takayasu arteritis with severe coronary, carotid, subclavian, and renal artery involvement and subsequent pregnancy. Interact Cardiovasc Thorac Surg 2006; 5: 153-5.
12. Kraemer B, Abele H, Hahn M, Rajab T, Kraemer E, Wallweiner D, et al. A successful pregnancy in a patient with Takayasu's arteritis. Hypertens Pregnancy 2008; 27: 247-52.
13. Matsumura A, Moriwaki R, Numano F. Pregnancy in Takayasu arteritis from the view of internal medicine. Heart Vessels Suppl 1992; 7: 120-4.

โรคทากายาสุในสตรีครรภ์แฝด: รายงานผู้ป่วย

จิตติมา รุจิเวชพงศธร, ยุรี ยานาเซะ

โรคทากายาสุเป็นโรคที่มีการอักเสบเรื้อรังของหลอดเลือดขนาดใหญ่ของร่างกาย ซึ่งการตั้งครรภ์ในรายที่มีโรคทากายาสุนั้นทำให้เกิดความเสี่ยงสูง วัตถุประสงค์ของรายงานผู้ป่วยนี้เพื่อบรรยายประสบการณ์ความสำเร็จของการดูแลสตรีตั้งครรภ์แฝดที่เป็นโรคทากายาสุ ผู้ป่วยรายนี้เป็นสตรีตั้งครรภ์อายุ 33 ปี ตั้งครรภ์ครั้งที่ 3 เคยแท้ง 1 ครั้ง และคลอดก่อนกำหนด 1 ครั้ง ได้รับการวินิจฉัยเป็นโรคทากายาสุตั้งแต่อายุ 15 ปี และได้รับการผ่าตัดหลอดเลือดใหญ่โป่งพองก่อนตั้งครรภ์ การตั้งครรภ์ครั้งนี้เป็นครรภ์แฝดและได้รับการดูแลโดยแพทย์สหสาขาที่คลินิกฝากครรภ์ความเสี่ยงสูง พบว่าทารกเสียชีวิต 1 คน เมื่ออายุครรภ์ 20 สัปดาห์ และทารกอีกคนมีการเจริญเติบโตช้าในครรภ์เมื่ออายุครรภ์ 25 สัปดาห์ สตรีครรภ์แฝดนี้ได้รับการผ่าตัดคลอดบุตรเมื่ออายุครรภ์ 30 สัปดาห์ เนื่องจากทารกมีการเจริญเติบโตช้า, มีความผิดปกติของการไหลเวียนในหลอดเลือดร่างกายทารก และภาวะครรภ์เป็นพิษ ได้ทารกเพศหญิงสุขภาพแข็งแรง น้ำหนักแรกคลอด 960 กรัม สรุปรายงานผู้ป่วยนี้แสดงให้เห็นถึงการดูแลสตรีตั้งครรภ์แฝดที่เป็นโรคทากายาสุที่สามารถดำเนินการตั้งครรภ์ได้อย่างปลอดภัยและคลอดบุตรมีชีวิตรอด แม้จะคลอดก่อนกำหนดร่วมกับโตช้าในครรภ์และมีภาวะแทรกซ้อนทารกเสียชีวิต 1 คน