CASE REPORT

Reversal of TTTS after fetoscopic laser photocoagulation for communicating vessels

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Accepted 25 March 2014

SUMMARY

We report a case of reversal of twin-to-twin transfusion syndrome (Stage IV TTTS) at 24 weeks of gestation after fetoscopic laser photocoagulation (FLP) for communicating vessels. The pathology of reversal of TTTS found that the 'initial' donor developed polyhydramnios whereas the 'initial' recipient developed oligohydramnios. With careful follow-up, the hydrops fetalis of the 'initial' recipient improved at 31 weeks of gestation. Because of the onset of labour pain at 34 weeks of gestation, two live and healthy babies were delivered by emergency caesarean section.

BACKGROUND

Twin-to-twin transfusion syndrome (TTTS) complicates approximately 10–15% of monochorionic, diamniotic twin pregnancies and is caused by an imbalance of placental blood flow with vascular anastomosis on the surface of the placenta.^{1–4} It is diagnosed by an ultrasound (US) examination showing the presence of polyhydramnios in the sac of one twin and oligohydramnios in the sac of the other. The donor fetus develops circulatory failure, anaemia, hypotension, oligouria, oligohydramnios, fetal growth retardation and renal failure because of chronic supply of blood flow. At the same time, the recipient fetus develops polyhaemia, hypertension, polyhydramnios, cardiac dysfunction and hydrops fetalis because of a circulatory overload. In severe cases, both fetuses are stillbirths. The prognosis is very poor in cases of onset before 26 weeks of gestation. In these cases, considering perinatal survival rates and long-term neurodevelopmental morbidities between 16 and 25 weeks of gestation, fetoscopic laser photocoagulation (FLP) for communicating vessels is advisable rather than amnioreduction.^{1–6} The procedures of FLP are to coagulate all anastomoses (artery to artery, venous to venous and artery to venous) by fetoscopic laser photocoagulation, after identifying anastomosis on the placental surface.^{1–4} As a result, the circulation in both fetuses becomes completely independent. If FLP is successful, TTTS improves after 1–2 weeks. Treatment outcomes of FLP in Japan showed an 80% survival rate, a 5% neurological sequelae rate and a 4% abortion rate.^{1–4} Two cases of reversal of TTTS (reversal of the donor–recipient phenotype) after FLP have been reported so far.^{7,8} The pathophysiology, incidence and optimal treatment remain to be clarified.

We present a case of Stage IV TTTS diagnosed at 24 weeks of gestation, followed by the reversal of TTTS after FLP. The reversed condition gradually

improved to a normal balance of both amniotic fluid volumes. As a result, the two babies were delivered by caesarean section at 34 weeks, followed by good neonatal outcomes.

CASE PRESENTATION

A 26-year-old multiparous woman was referred to our hospital for perinatal care of a monochorionic diamniotic twin pregnancy at 15 weeks and 3 days of gestation. US examinations were started and performed every 2 weeks thereafter. At 22 week and 0 day the US examination showed no problem between the two estimated fetal body weights (EFBW) (492 vs 517 g). Also, no structural abnormalities in the two fetuses were detected, and the maximum vertical pocket (MVP) of the amniotic cavity were normal in both (4.1 vs 6.0 cm) (figure 1). However, as shown in figure 1, the MVPs at 24 weeks and 0 day were distinctly different between the twins (twin A/initial recipient; 8.3 cm vs twin B/initial donor 1.4 cm), along with the hydropic change of ascites in twin A (initial recipient). Therefore, the patient was diagnosed in Quintero Stage IV TTTS and FLP was indicated as one of the treatment options. After obtaining an informed consent for FLP, the patient was transferred to Tokuyama Central Hospital at 24 weeks and 3 days for the procedure to be performed. US examination showed typical signs such as the hydropic changes of subcutaneous oedema, ascites and pericardial effusion, and also reversed flow during atrial contraction in the ductus venosus in twin A (initial recipient), which were characteristic of hydrops fetalis with congestive heart failure. During FLP at 24 weeks and 3 days, all anastomosis were photocoagulated. The arteriovenous (AV) anastomosis consisted of one anastomosis from twin A (initial recipient) to twin B (initial donor), three anastomoses from twin B (initial donor) to twin A (initial recipient) and an arterio–arterio (AA) anastomosis was recognised from twin B (initial donor) to twin A (initial recipient).

One day after FLP, US examination demonstrated a voiding and an increase of amniotic fluid volume in twin B (initial donor). However, 7 days after FLP the amniotic fluid imbalance (polyhydramnios/oligohydramnios) in twin B (initial donor) had not been corrected. Therefore, 8 days after FLP (25 weeks and 4 days) the amnioreduction of 1000 mL was carried out in twin B (initial donor). Meanwhile, 9 days after FLP the amniotic fluid of twin A (initial recipient) increased and a reversed flow during atrial contraction in the ductus venosus was observed again in twin A (initial recipient).

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To cite: Miyazaki M, Honda N, Yokoyama M, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-202711

Figure 1 Time course of most vertical amniopocket (MVP) displayed hydrops fetalis of twin A (initial recipient; solid line) at 24 weeks and 0 day, and displayed hydrops fetalis of twin B (initial donor; dotted line) at 26 weeks and 4 days. Reversal of maximum vertical pocket (MVP) was recognised at 26 weeks and 6 days. Reversal of twin-to-twin transfusion syndrome (TTTS) was diagnosed at 28 weeks and 0 day (FLP, fetoscopic laser photocoagulation).

Although a second FLP was planned to be conducted, 10 days after FLP (25 weeks and 6 days) a high rupture of the bag of waters occurred.

Because of a higher risk of premature deliveries in the case of deterioration of TTTS, the patient was transferred back to our hospital near her residence at 26 weeks and 0 day. US examinations showed twin B (initial donor) with hydrops fetalis at 26 weeks and 3 days, and the MVPs of both the twins had completely reversed at 26 weeks and 5 days, that is, twin A: initial recipient, new donor/1.5 cm and twin B: initial donor, new recipient/8 cm. Accordingly, the patient was diagnosed with a reversal of TTTS after FLP.

As a very high-risk case of the reversal of TTTS after FLP, careful monitoring was continued of the fetal conditions with US examinations and cardiotocograms. The parameters evaluated were the Doppler velocity wave forms of both the fetal mid-cerebral arteries (MCA), the umbilical arteries (UMA) and both MVPs of amniotic fluids.

Doppler velocity waveforms of MCA and UMA maintained average levels throughout the observation periods (figure 2).

The assessment of the fetal cardiac function was performed by preload index (PLI) and cardiothoracic area ratio (CTAR) (figure 3). The PLI and CTAR of twin A (initial recipient, new donor) were high through the first 28 weeks of gestation, but gradually decreased to within the normal range. US examinations documented a gradual increase of amniotic fluid in twin A (initial recipient, new donor) and a gradual decrease of amniotic fluid in twin B (initial donor, new recipient) after 29 weeks and 1 day. The MVPs of both fetuses were in the normal range from 29 weeks, and were similar from 30 weeks (figure 1). The ascites of twin B (initial donor, new recipient) disappeared at 29 weeks and 1 day, and the ascites of twin A (initial recipient, new donor) dissolved at 31 weeks and 1 day (figure 4).

The discordance of EFBW was 27% at 26 weeks and 4 days, but gradually declined to 0.8% after 29 weeks and 1 day (figure 5).

The patient delivered the babies by emergency caesarian section at 34 weeks and 5 days, due to uncontrollable labour pain. Twin A (initial recipient, new donor) was a 2190 g female infant with an Apgar score of 6 and 8 at 1 and 5 min,

RI-UmA, RI-MCA

Figure 2 Changes in resistance index—umbilical artery (RI-UmA) and resistance index—middle cerebral artery (RI-MCA) of twin A (initial recipient; solid line) and twin B (initial donor; dotted line). Daily ultrasound examinations displayed the main reference values throughout pregnancy.

respectively. The pH of the umbilical artery was 7.294. Twin B (initial donor, new recipient) was a 2114 g female infant with an Apgar score of 8 and 8 at 1 and 5 min, respectively. The pH of the umbilical artery was 7.264 with a good neonatal outcome. Both infants were discharged from the neonatal intensive care unit (NICU) of our hospital on postnatal day 36 with no neurological abnormality and with average growth.

INVESTIGATIONS

US examinations: estimated fetal body weights (EFBW), most vertical amniopocket (MVP), resistance index—umbilical artery (RI-UmA), resistance index—middle cerebral artery (RI-MCA), preload index (PLI), cardiothoracic area ratio (CTAR), Cardiotocograph (CTG).

TREATMENT

- ▶ Fetoscopic laser photocoagulation (FLP) for communicating vessels
- ▶ Careful follow-up with the US examinations

Figure 3 The assessment of the fetal cardiac function was performed by preload index (PLI) and cardiothoracic area ratio (CTAR). Twin A (initial recipient; solid line) is initial recipient with gradually improved cardiac function. PLI: reference value <0.5, CTAR: reference value: 25–35% (TTTS, twin-to-twin transfusion syndrome).

Figure 4 Ultrasound examination showed ascites in the twins at 26 weeks. However, the ascites in the twins dissolved at 31 weeks.

OUTCOME AND FOLLOW-UP

Both infants discharged from the NICU of our hospital at post-natal day 36, with no neurological abnormalities and with average growth.

DISCUSSION

The aetiology of TTTS is attributed to the presence of anastomosis on the placenta between the two fetal circulations, resulting in acute hemodynamic changes. There is growing evidence indicating that FLP is the most effective therapy to treat TTTS.^{1–3}

The pathology of the reversal of TTTS relies on reversing the donor–recipient phenotype, that is, a donor fetus acquiring features of a recipient and vice versa. Several studies of the reversal of TTTS have been reported recently.^{7–9} However, the reversal of TTTS after FLP as described here was reported in only two cases.^{7,8} The pathologies, the incidence rates and the treatment protocols of the reversal of TTTS remain to be determined.

In the two case reports of the reversal of TTTS after FLP, one documented the reversal of TTTS at 6 h after FLP and both twins died in utero.⁸ The other described type of reversal of TTTS occurred at 7 weeks after FLP, resulting in surviving twins.⁷ In our case, the reversal of amniotic fluid of donor–recipient gradually developed 2 weeks after FLP. The reversed

hemodynamics was subsequently ameliorated without additional FLP or other treatment. Placental study with injection of coloured dye confirmed no residual anastomosis on the surface of the placenta. The fractional shortening (FS) and Vmax on another assessment of the fetal cardiac function, of twin B (initial donor, new recipient) had lower than normal values in 26 weeks, but improved in 30 weeks (data not shown). Therefore, it could be assumed that the reversal of TTTS in our case was due to twin B (initial donor, new recipient) developing relative heart failure under the improved hemodynamics by FLP. The reversal of TTTS after FLP is considered to occur during the course of normalisation of the blood flow between the fetuses. Consequently, the reversal of TTTS might spontaneously improve depending on the cardiac function of the initial donor/new recipient. Since the curative effect of FLP may be continuously exerted in the course of the twin pregnancy, a careful follow-up for the changes of hemodynamics between the twins is necessary.

Learning points

- Fetoscopic laser photocoagulation for communicating vessels (FLP) is a curative treatment for severe twin-to-twin transfusion syndrome (TTTS) between 16 and 25 weeks of gestation.
- Although the reversal of TTTS occurs very rarely after FLP, the potential for its occurrence after the treatment should be considered.
- Since the curative effect of FLP may be continuously exerted in the course of the twin pregnancy, a careful follow-up is necessary for the hemodynamic changes between the twins.

Acknowledgements The authors would like to acknowledge the obstetricians of Tokuyama Central Hospital and the paediatricians of Matsuyama Red Cross Hospital for the treatment of the twins.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Murakoshi T. Current status of prenatal treatment(2); laser-treatment of multiple pregnancy. *Matern Child Health Inf* 2010;61:40–5.
- 2 Murakoshi T. Management of twin-twin transfusion syndrome. *Japan Soc Obstet Gynecol* 2007;59:588–95.
- 3 Murakoshi T. Management of abnormal twin pregnancy; pathophysiology and treatment for twin-twin transfusion syndrome. *Japan Soc Obstet Gynecol* 2003;55:216–23.
- 4 Hayashi S. Investigation of 25 cases of twin-to-twin transfusion syndrome underwent fetoscopic laser therapy. *Bull Kanto Japan Soc Obstet Gynecol* 2006;43:413–18.
- 5 Middeldorp JM, Lopriore E, Suetters M, et al. Twin-to-twin transfusion syndrome after 26 weeks of gestation: is there a role for fetoscopic laser surgery? *Br J Obstet Gynaecol* 2007;114:694–8.
- 6 Leullo AM, Papageorgiou AT, Bhida A, et al. Severe twin-twin transfusion syndrome: outcome after fetoscopic laser ablation of the placental vascular equator. *Br J Obstet Gynaecol* 2007;114:689–93.
- 7 Fox CE, Chan BC, Cox P, et al. Reverse twin-to-twin transfusion syndrome following successful laser therapy. *Fetal Diagn Ther* 2009;26:115–18.
- 8 Wataganara T, Chanprapaph P, Chuangsuwanich T, et al. Reverse twin-twin transfusion syndrome after fetoscopic laser photocoagulation of chorionic anastomoses: a case report. *Fetal Diagn Ther* 2009;26:111–14.
- 9 Sumie M, Nakata M, Murata S, et al. Two cases of reversal of twin-twin transfusion syndrome diagnosed by measuring hourly fetal urine production. *J Obstet Gynaecol Res* 2009;35:983–6.

Figure 5 The estimated fetal body weights (EFBW) of the twins (twin A; solid line and twin B; dotted line) displayed no discordancy after 29 weeks (TTTS, twin-to-twin transfusion syndrome).

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