CASE REPORT

Severe neonatal complication of transverse lie after preterm premature rupture of membranes

D C M Van der Kaay,¹ S Horsch,¹ J J Duvekot²

¹Department of Pediatrics, Erasmus MC, Rotterdam, Netherlands
²Department of Obstetrics, Erasmus MC, Rotterdam, Netherlands

Correspondence to
 Dr Johannes Duvekot,
 j.j.duvekot@erasmusmc.nl

SUMMARY

Both transverse lie and preterm premature rupture of membranes (PPROM) are associated with neonatal morbidity and mortality. We present a neonate born at 29 weeks gestation with severe birth trauma after PPRM and transverse lie. The patient had extensive swelling and areas of desquamated and necrotic skin of the right lower limb. Neonatal compartment syndrome (NCS) was suspected. Perfusion of the limb improved after decompressing subcutaneous incisions. A fetus in transverse lie may be mechanically damaged in the case of PPRM, especially at an early gestational age. Early recognition is of great interest in the management and prognosis of NCS.

BACKGROUND

Preterm birth accounts for 5–10% of all deliveries and preterm premature rupture of membranes (PPROM) accounts for one-third of those preterm deliveries.¹ An important complication of PPRM is fetal non-vertex presentation at delivery.

The proportion of fetus with non-vertex presentation decreases with increasing gestational age (28% at 25–28 weeks gestation; 2% at 37–40 weeks gestation).² The most common non-vertex presentation in preterm births is breech position (75%), followed by transverse lie (21%) and other presentations including compound position (4%).

We present a preterm neonate who was born with severe birth trauma after PPRM and transverse lie. The neonate was born after an emergency caesarean section because of fetal distress. At birth, there was extensive swelling and areas of desquamated and necrotic skin of the right lower limb.

CASE PRESENTATION

A female macrosomic neonate (birth weight 1525 g) was born preterm at 29 weeks gestation in a nulliparous mother whose pregnancy was complicated by threatened preterm labour earlier in pregnancy. The mother had facioscapulohumeral muscular dystrophy. Chorion biopsy did not reveal the same gene defect in the fetus. After PPRM resulting in oligohydramnios at 28 weeks and 3 days, the fetus presented in transverse lie with the fetal back upwards. The cervix was closed during internal examination. Initially, expectant management was instituted until 4 days after admission an emergency caesarean section was performed because of the suspicion of fetal distress because of decelerations on the cardiotocogram. There were no signs of labour at that time.

At delivery, the fetus still presented in transverse lie. The neonate could be delivered in breech position with some difficulty. Apgar scores after 1 and 5 min were 5 and 7, respectively. Because of persistent respiratory distress, the neonate was intubated 20 min after delivery. Owing to a subgaleal bleeding accompanied by hypotension and tachycardia, she received saline, fresh frozen plasma and two blood transfusions in the first few hours after delivery.

At birth, extensive swelling in areas of desquamated and necrotic skin of the right lower limb was noted (figure 1).

INVESTIGATIONS

Full blood count was within the normal range. A blood culture demonstrated an infection with *Streptococcus anginosus* for which she was treated with penicillin during 10 days. C reactive protein levels increased to a maximum of 7 mg/L. X-rays of both lower limbs did not demonstrate fractures or bone deformities. Although flow in the femoral and tibial arteries was detectable, capillary refill in the right toes was diminished. Doppler ultrasound suggested raised pressure in the arterial lower limb trajectory without signs of thrombosis.

TREATMENT

Because of extensive subcutaneous oedema at the dorsal side of the right foot, decompressing subcutaneous incisions were performed. The perfusion of the toes improved. Emergency fasciotomy was not necessary.

OUTCOME AND FOLLOW-UP

In the following days, the extensive swelling decreased and the skin lesions recovered (figure 2).

Figure 1 Extensive swelling with areas of desquamated and necrotic skin of the right lower limb immediately after birth.

To cite: Van der Kaay DCM, Horsch S, Duvekot JJ. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2012-008399

Figure 2 Extensive swelling decreased after decompressing subcutaneous incisions, and the skin lesions recovered after a couple of days. Note the equinus position of the right foot with plantar flexion of the toes.

The right lower limb lacked spontaneous movement and the neonate developed an equinus position of the foot with plantar flexion of the toes, for which a paediatric orthopaedist was consulted at day 7. Physical therapy was started in order to maintain maximal range of motion. She was transferred to a high care centre after 12 days, where she was discharged home after 5 weeks. Physical therapy was continued at home. The neonate was seen at the outpatient orthopaedic clinic at a corrected 40 weeks of gestation. The aspect and functionality of the right limb was completely normal and there were no signs of muscular atrophy.

DISCUSSION

We present a neonate born after PPRM and transverse lie at 29 weeks of gestation with signs of neonatal compartment syndrome (NCS). The neonate was born after an emergency caesarean section because of the suspicion of fetal distress. To our knowledge, this is the first presentation of a prematurely born neonate with signs of NCS of the right lower limb after PPRM and transverse lie.

An important complication of PPRM is fetal malpresentation at delivery since spontaneous version with PPRM is unlikely.³ Complications such as placental abruption, chorioamnionitis, umbilical cord prolapse and direct birth trauma are more common and perinatal mortality is higher in non-vertex presentation compared with vertex presentation.² Transverse lie with the mother in labour or with ruptured membranes was associated with lower arterial pH (7.21 compared with 7.26 in breech and vertex group), more frequent severe acidosis (pH<7.10: 21% compared with 0% in breech and vertex group) and more frequent birth trauma such as brachial plexus palsy and extensive ecchymosis (overall morbidity 36% compared with 4% in breech respectively 7% in vertex group).⁴

Management of the fetus in transverse lie depends upon the clinical circumstances at the time of diagnosis. Length of gestation and the intactness of the membranes are important factors to consider. Caesarean section was found to have a protective effect on neonatal outcome and is generally recommended if the mother is in labour or membranes have ruptured.² However, it is difficult to choose the exact time for the caesarean section once the diagnosis of transverse lie and PPRM at an early gestational age is established. In our patient, PPRM was noted 4 days before an emergency caesarean section was performed

because of suspected fetal distress. The severe neonatal morbidity could possibly have been prevented if a caesarean section was performed earlier.

The exact pathological mechanism of NCS is unknown. Extrinsic causes include mechanical compression, amniotic band constriction or direct birth trauma. Intrinsic causes include hypercoagulable state of the neonate leading to arterial or venous thrombosis. Both extrinsic and intrinsic causes lead to increased intracompartmental pressure resulting in ischaemia with muscle and nerve injury.⁵ Predisposing factors for NCS are preterm delivery, oligohydramnios and an abnormal fetal position, maternal diabetes, polycythaemia, neonatal respiratory distress, difficult vaginal delivery and multiple gestation. In our patient, oligohydramnios in combination with transverse position could have led to intrauterine compression of the right lower limb.

Presenting symptoms of NCS include bullous or ulcerative skin lesions with distal oedema. Upper extremities—typically the dorsal forearm, wrist and/or hand—are almost exclusively involved. Lesions vary from a blue plaque, local ulceration, eschar and bullae to gangrene. Pulses are usually palpable, whereas capillary refill can be diminished. Diagnostic tools include serial photographs, Doppler ultrasounds and MRI. Management includes fasciotomy if blood flow is compromised. In neonates presenting with extensive necrosis or severe contractures without oedema, fasciotomy is probably futile. In these patients, surgical debridement and skin grafts are often necessary because of underlying necrosis.^{5 6} In our patient, fasciotomy was not performed as perfusion of the toes improved after decompressing subcutaneous incisions. We feel that the decompressing incisions contributed to the favourable outcome. However, perfusion might also have improved spontaneously, so this case illustrates how difficult diagnosis and treatment are in a preterm neonate. Long-term outcome is variable, depending on the extent of damage and possible delay in diagnosis and treatment, resulting in contractures, loss of function, neuropathy and amputation. Fortunately, in our patient, functionality of the right limb had normalised at term equivalent age.

NCS is a rare and an initially often misdiagnosed condition because the skin lesions mimic other diseases of the newborn such as necrotising fasciitis and neonatal gangrene. Necrotising fasciitis (NF) should be considered if blood or tissue cultures are positive. NF is associated with rapid clinical deterioration, followed by death in a large part of newborns.⁷ In our patient, a blood culture demonstrated *S. anginosus* for which she received penicillin for 10 days. These bacteria are part of the human bacterial flora and can cause a bacteraemia after local trauma to the mucosal barrier, for example, after intubation. The clinical course in our patient was not consistent with the course of NF. Neonatal gangrene is more commonly found in the lower extremity and has more serious complications requiring amputations. Risk factors are similar to those for NCS. Arterial pulses are usually absent in the affected limb.⁸

In conclusion, we present a neonate born after PPRM and transverse lie at 29 weeks gestation with signs of NCS after an emergency caesarean section. Especially in cases of oligohydramnios, prolonged transverse lie may harm the fetus. The timing of performing a caesarean section can be very difficult and it is thus important to closely monitor the unborn neonate in order to prevent complications. Early recognition and subsequent choice of treatment of neonatal compartment syndrome can be very challenging, but is of great interest in the management and prognosis.

Learning points

- ▶ A fetus in transverse lie may be mechanically damaged in case of preterm premature rupture of membranes (PPROM), especially at an early gestational age.
- ▶ Once the diagnosis of transverse lie and PPRM at an early gestational age is established, the fetus should be monitored meticulously/carefully.
- ▶ Although upper extremities are almost exclusively involved in neonatal compartment syndrome, we presented a neonate with signs of neonatal compartment syndrome (NCS) at the lower extremity.
- ▶ Early recognition and choice of treatment of NCS can be challenging, but is of great interest in the management and prognosis.

Contributors DCMVdK and SH had the idea for the article. All three authors performed the literature search. DCMVdK wrote the article. JD is the guarantor.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Mercer BM. Preterm premature rupture of the membranes: diagnosis and management. *Clin Perinatol* 2004;31:765–82, vi.
- 2 Demol S, Bashiri A, Furman B, *et al*. Breech presentation is a risk factor for intrapartum and neonatal death in preterm delivery. *Eur J Obstet Gynecol Reprod Biol* 2000;93:47–51.
- 3 Fonseca L, Monga M. Spontaneous version following preterm premature rupture of membranes. *Am J Perinatol* 2006;23:201–3.
- 4 Hankins GD, Hammond TL, Snyder RR, *et al*. Transverse lie. *Am J Perinatol* 1990;7:66–70.
- 5 Ragland R III, Moukoko D, Ezaki M, *et al*. Forearm compartment syndrome in the newborn: report of 24 cases. *J Hand Surg Am* 2005;30:997–1003.
- 6 Allen LM, Benacci JC, Trane RN III, *et al*. A case of neonatal compartment syndrome: importance of early diagnosis in a rare and debilitating condition. *Am J Perinatol* 2010;27:103–6.
- 7 Peker E, Kirimi E, Tuncer O, *et al*. Necrotizing fasciitis caused by *Staphylococcus epidermidis* in a neonate with extremely low birthweight. *J Dermatol* 2010;37:671–3.
- 8 Nagai MK, Littleton AG, Gabos PG. Intrauterine gangrene of the lower extremity in the newborn: a report of two cases. *J Pediatr Orthop* 2007;27:499–503.

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow