CASE REPORT

Subdural haemorrhage and severe coagulopathy resulting in transtentorial uncal herniation in a neonate undergoing therapeutic hypothermia

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SUMMARY

Therapeutic hypothermia has been shown to be efficacious for improving long-term neurodevelopmental outcomes following perinatal asphyxia. Thus, cooling protocols have been adopted at most tertiary neonatal centres. We present a case of a term neonate who underwent therapeutic whole-body cooling for hypoxic ischaemic encephalopathy following a difficult forceps delivery. She abruptly deteriorated, exhibiting signs of transtentorial uncal herniation and severe disseminated intravascular coagulopathy. CT of the head confirmed a life-threatening subdural haematoma and a concealed skull fracture. Hypothermia has been shown to impair haemostasis in vivo and thus may potentially exacerbate occult haemorrhages in a clinical setting. Newborns that require instrument-assisted delivery are a particularly high-risk group for occult head injuries and should undergo careful clinical assessment for fractures and intracranial haemorrhage prior to initiation of therapeutic hypothermia.

BACKGROUND

Perinatal asphyxia encephalopathy or hypoxic ischaemic encephalopathy (HIE) is an important cause of death and severe disability in childhood. In the past decade, numerous multicentre randomised control trials have demonstrated long-term neurodevelopmental benefits of hypothermia in the management of moderate perinatal HIE.¹ The adverse events reported in these studies were generally minor and/or not associated with cooling. As such, whole-body hypothermia has been widely adopted into clinical practice. However, as increasing numbers of babies are undergoing this treatment, the safety of this practice needs to be monitored and serious adverse events during this therapy should be reported. This is the first case in the literature of a neonate undergoing therapeutic hypothermia who subsequently developed a massive subdural haemorrhage and concurrent disseminated intravascular coagulopathy (DIC) resulting in transtentorial uncal herniation.

CASE PRESENTATION

A 29-year-old previously healthy woman presented in spontaneous labour at 39+2 weeks gestation. She had antenatal ultrasounds documenting a breech presentation and normal fetal anatomy. Prior to delivery, the fetus was noted to have variable decelerations with heart rate dropping to the 60 s. During delivery, head extraction was

extremely difficult requiring suprapubic pressure and forceps assistance.

The infant weighed 2.718 kg with APGARs of 0¹, 0⁵, 2¹⁰, 5¹⁵, 5²⁰. Her cord pH was 6.99 and base deficit was -13.8. During resuscitation she required intubation, chest compressions and two doses of intravenous epinephrine. Her first gasp and heart rate were recorded at 11 min. She was hypotonic with minimal spontaneous movements. Passive cooling was initiated at 30 min of life. Her temperature reached 34°C rectal at 70 min, which is in the target range of 33-34°C rectal for therapeutic hypothermia.

On transfer to our tertiary care centre she was noted to have a stable posterior subgaleal haematoma with multiple skin abrasions and forceps markings over her head and scalp. However, there was no bony crepitus or deformities of the skull. Her pupils were equal and reactive. She had a weak cry, suck and gag, with a partial moro reflex and bilateral absence of her grasp reflexes. She began spontaneously breathing with minimal ventilator settings and was subsequently extubated to room air on arrival to our centre and continued on our hypothermia protocol.

INVESTIGATIONS

The infant's first complete blood count in the referring hospital showed a leucocyte count of $19.6 \times 10^6/L$, platelet count of $187 \times 10^9/L$ and haemoglobin of 169 g/L. On transfer to our tertiary care centre, this was repeated and found to be stable with platelets of $203 \times 10^9/L$ and haemoglobin of 142 g/L.

Owing to the difficult delivery and the presence of a subgaleal haemorrhage, skull X-ray was obtained and showed a very mildly depressed fracture of the occipital bone (figure 1). The baby was subsequently sent for a CT scan of the head (figure 2) which revealed a large right side subdural haemorrhage exerting mass effect and causing an 8 mm midline shift. Crowding around the right tentorium cerebelli was noted. A left subgaleal haemorrhage was also present with an underlying 'ping-pong' type skull fracture with a maximum depression of 3 mm.

TREATMENT

Immediately after her CT scan at 14 h of life, the baby developed periodic breathing and was found to have a fixed and dilated right pupil. She was reintubated and active rewarming was initiated. She received treatment for increased intracranial

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Figure 1 Skull X-ray showing a minimally depressed skull fracture in the posterior left parietal bone just superior to the lambdoid suture.

pressure and neurosurgery was consulted for emergency decompression of the right subdural haematoma. Concurrently, her laboratory results were suggestive of DIC with a haemoglobin of 94 g/L, platelet count of $82 \times 10^9/\text{L}$, international normalised ratio (INR) of 2.7, partial thromboplastin time (PTT) of 42 s and fibrinogen of 1.17 g/L. She had evidence of mucosal and gastric bleeding.

She received large volumes of blood product resuscitation and required a total of 10 mL/kg of fresh frozen plasma, 40 mL/kg of cryoprecipitate, 15 mL/kg of platelets, 15 mL/kg of packed red blood cells and 1 mg intravenous vitamin K. Haematology was consulted and suggested a dose of factor VII at 90 mcg/kg but her coagulopathy corrected prior to administration with laboratory results showing an INR of 1.4, PTT 30.5 s, fibrinogen 2.64 g/L, haemoglobin of 79 g/L and platelet of 288 000. Her temperature at this time had also normalised to 35°C axillary.

Once her coagulopathy improved, a right decompressing craniectomy was performed and a 200 mL clot was evacuated.

OUTCOME AND FOLLOW-UP

Postoperative CT of the head showed resolution of the midline shift. She recovered quickly and the remainder of her hospital stay was unremarkable. She was discharged home on day 16 of life with some mild residual left-sided weakness. Coagulation profiles and platelet count were completely normal at the time of discharge.

At her 18-month follow-up she was developmentally normal. She had no seizures. Her neurological exam was unremarkable. She had a persistent skull deformity secondary to the healing depressed skull fracture and is awaiting surgical replacement of the bone fragment from the craniectomy. No clinical concerns

of prolonged bleeding or easy bruising were present on follow-up visit.

DISCUSSION

Previously published cooling trials have found no statistically significant increase in the incidence of bleeding, coagulopathy or thrombocytopenia in the hypothermic groups. In a safety outcome analysis from the National Institute of Child Health and Human Development (NICHD) trial, there was no statistically significant increase in bleeding episodes.² However, the protocol did not investigate the coagulation profile or blood viscosity status for all enrolled patients. Furthermore, many of the studies including the NICHD trial² were not powered to assess for harm. Consequently, none of the published cooling trials have specifically addressed infants at risk of occult intracranial haemorrhage who also meet criteria for therapeutic hypothermia. There are no similar published cases in the literature of a neonate undergoing cooling who subsequently developed a massive subdural haemorrhage and concurrent DIC resulting in transtentorial uncal herniation.

Numerous published reports have shown that the risk of intracranial injury is significantly increased in instrumented deliveries. The risk of an associated intracranial haemorrhage is shown in one study to increase from 0% to 28% for instrument-associated depressed skull fractures versus spontaneous ones.³ With the increased likelihood of intracranial bleeding, it is reasonable to also consider the effect that hypothermia can have on haemostasis in neonates requiring an instrument delivery who also meet criteria for cooling. In vivo studies have shown that hypothermia can cause coagulation abnormalities such as platelet dysfunction and increased fibrinolytic activity. There is also inhibition of enzymatic reactions of the

Figure 2 CT of the head (axial and coronal views) revealed a right subdural haematoma exerting mass effect and causing an 8 mm midline shift. Crowding around the right tentorium cerebelli was also noted (not shown). Left subgaleal haemorrhage was seen with an underlying 'ping-pong' skull fracture with a maximum depression of 3 mm.

coagulation cascade with prolongation of prothrombin time and partial thromboplastin time.⁴ In addition, an updated Cochrane review of cooling trials to date showed a statistically significant increase in thrombocytopenia in hypothermic groups.¹ There is also a trend towards a significant increased risk of coagulopathy including a higher incidence of coagulopathy resulting in major thrombosis or haemorrhage in the hypothermia group.¹ The degree of bleeding dysfunction appears to be associated with the severity of hypothermia.⁵ Temperatures below 35°C can cause platelet dysfunction and a decrease in platelet count.⁵ At temperatures less than 33°C, the synthesis and kinetics of the clotting cascade are also affected.⁵ While the risk of spontaneous bleeds does not appear to be increased,^{4 5} adult studies on hypothermia suggest that those developing bleeding complications while hypothermic require more red blood cell unit transfusions to achieve haematological stability.⁵

Although significant asphyxia can also impair coagulation function,⁶ we hypothesise that therapeutic hypothermia played a contributory role in this case as the patient's coagulopathy was

unusually severe and required extensive amounts of blood product administration and rewarming before stabilisation. In addition, while small subdural haematomas are common in neonates born via instrumented deliveries,⁷ transtentorial uncal herniation is extremely rare in newborns and only occurs in situations with a rapid, acute rise in intracranial pressure.⁸ In our case this was secondary to massive intracranial bleeding. Subdural haemorrhages of this severity in newborns are often associated with pathology such as an underlying coagulopathy.^{9 10}

Contributors DW drafted the manuscript. HM provided images, reviewed and revised the manuscript. EB reviewed and revised the manuscript.

Competing interests None.

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Learning points

- The safety of therapeutic hypothermia requires ongoing surveillance and evaluation.
- Careful and thorough evaluation of the clinical circumstances should always be performed when selecting patients for therapeutic hypothermia.
- Neonates requiring instrument deliveries are at risk of occult intracranial bleeds and careful clinical evaluation should be performed with a low threshold for cerebral imaging.
- Neonates requiring instrument deliveries who meet criteria for therapeutic hypothermia should be closely monitored for the development of coagulation abnormalities and worsening intracranial bleeding while undergoing cooling.

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