


# Unsupervised breastfeeding was related to sudden unexpected postnatal collapse during early skin-to-skin contact in cerebral palsy cases

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## Abstract

**Aim:** This study aimed to identify the clinical features of infants who were healthy at birth, but developed sudden unexpected collapse and were then diagnosed with cerebral palsy before 5 years of age.

**Methods:** We retrospectively analysed 1182 records from the no-fault Japan Obstetric Compensation System for Cerebral Palsy database up to 2016. This identified 45 subjects (3.8%) who were subsequently diagnosed with severe cerebral palsy due to sudden unexpected postnatal collapse (SUPC). They were all healthy at birth, based on the criteria of five-minute Apgar scores of seven or more, with normal umbilical cord blood gases and no need for neonatal resuscitation within five minutes of birth.

**Results:** The median birth weight of the 45 subjects (26 males) was 2770 g (range 2006–3695 g). Of these, 10 developed SUPC during early skin-to-skin contact (SSC). Medical personnel were not present in all 10 cases: nine were being breastfed at the time and eight of the mothers did not notice their infant's abnormal condition until medical staff alerted them.

**Conclusion:** This national study of children with cerebral palsy who appeared healthy at birth found that unsupervised breastfeeding was a common factor in cases of SUPC during early SSC.

## KEYWORDS

breastfeeding, cerebral palsy, early skin-to-skin contact, sudden unexpected postnatal collapse

**Abbreviations:** SSC, skin-to-skin contact; SUPC, sudden unexpected postnatal collapse.

\*These Prevention Recurrence Committee, Japan Obstetric Compensation System for Cerebral Palsy members present in Acknowledgements section.

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## 1 | INTRODUCTION

Dramatic advances in perinatal care have led to improved survival rates for high-risk neonates, including premature infants. Despite this, the incidence of cerebral palsy has been reported to be two per 1000 deliveries<sup>1-3</sup> and this has not changed in the last few decades. This means that cerebral palsy remains an important issue in perinatal medicine. Several causes of cerebral palsy have been identified, including preterm birth, hypoxic or ischaemic damage, congenital anomalies and infections. Generally, these infants show abnormal symptoms immediately after birth and are given neonatal resuscitation and intensive medical care. However, even apparently healthy infants who do not show abnormal symptoms at birth can have a sudden unexpected postnatal collapse (SUPC) during their postnatal stay in the maternity ward or neonatal nursery. They can then go on to be diagnosed with cerebral palsy.<sup>4</sup>

The Japan Obstetric Compensation System for Cerebral Palsy was established in 2009 to provide no-fault compensation for children with severe disabilities due to cerebral palsy as a result of trauma during labour and delivery. It does not cover congenital causes or factors during the neonatal period or later. The system also aims to prevent cerebral palsy, by achieving early resolution of medical conflicts and improving obstetric care. Disability is measured on a seven-degree scale laid down in the Japanese Act for the Welfare of Persons with Physical Disabilities. The system compensates children with a first or second degree disability, which corresponds to severe locomotor disabilities and specifically indicates that they won't be able to walk. Families contact the delivery facilities, and they apply to the Japan Council for Quality Health Care for certification examinations undertaken from the child's first to fifth birthday. Based on primary screening by paediatricians and obstetricians with expertise in cerebral palsy, cases undergo a secondary review by committee of paediatricians, psychiatrists, obstetricians and academic experts

### Key notes

- We used a national no-fault compensation database to study 45 children with cerebral palsy who were apparently born healthy, but then had a sudden unexpected postnatal collapse (SUPC).
- The data showed that 27 developed SUPC during rooming-in with their mothers and 10 of these collapsed during early skin-to-skin contact (SSC).
- Unsupervised breastfeeding was a common factor in the infants who had an SUPC during early SSC.

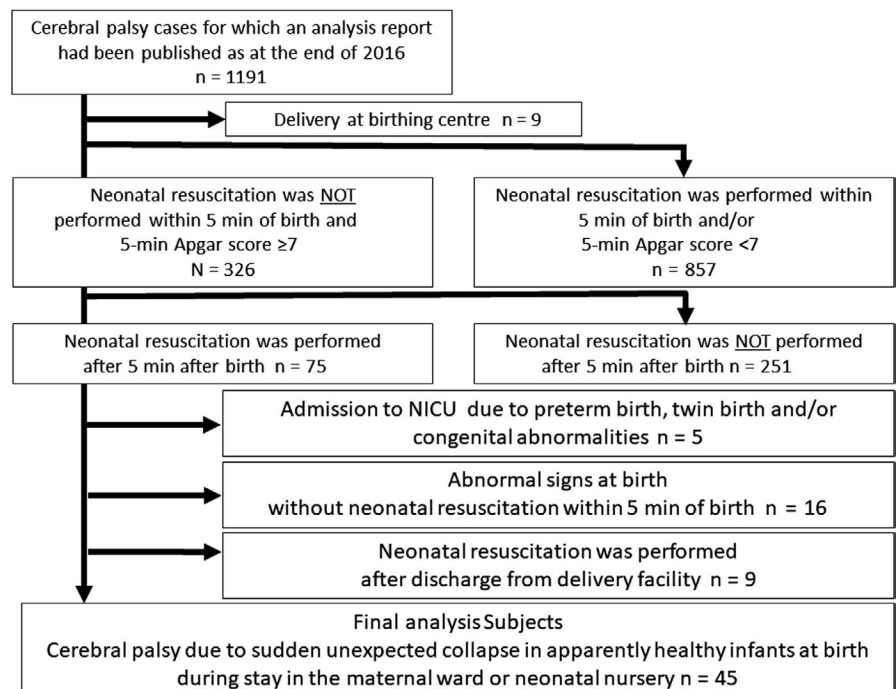
who approve or reject compensation claims. Approved cases are then analysed by obstetricians, paediatricians, midwives, lawyers and intellectuals, to identify the cause, and their report is sent to the delivery facility and families.<sup>4</sup>

Our aim was to identify the clinical features of infants who developed SUPC after they were deemed healthy at birth and subsequently developed cerebral palsy before 5 years of age. We did this by using the Japanese database to retrospectively review the children's clinical backgrounds, the situations at the onset of SUPC and pathophysiologic conditions that were presumed to be the main causes of cerebral palsy.

## 2 | PATIENTS AND METHODS

### 2.1 | Subjects

We studied infants with severe cerebral palsy due to SUPC during their stay in delivery facilities. All the cases we included had been



**FIGURE 1** Flow diagram showing the number of eligible cases

**TABLE 1** Clinical background and characteristics of the subjects

Variables	Cases (n = 45)
Maternal characteristics	
Age (years)	32 (20-45)
Primipara	33 (73.3%)
Multiple birth	4 (8.9%)
Pregnancy-induced hypertension	4 (8.9%)
Diabetes mellitus/gestational diabetes mellitus	1 (2.2%)
Premature rupture of membranes	9 (20.0%)
Clinical chorioamnionitis	0
At delivery	
Delivery facilities	
Hospitals	30 (66.7%)
Clinics	15 (33.3%)
Painless delivery	3 (6.7%)
Use of uterine contraction agents	17 (37.8%)
Placental abruption	2 (4.4%)
Fore-lying of umbilical cord	1 (2.2%)
Foetal heart rate monitoring abnormalities	27 (60%)
Meconium-stained amniotic fluid	10 (22.2%)
Acute delivery	
Instrumental (due to non-reassuring foetal status)	2 (4.4%)
Instrumental (due to prolonged delivery or maternal fatigue)	2 (4.4%)
Emergent Caesarean section (due to non-reassuring foetal status)	2 (4.4%)
Emergent Caesarean section (due to prolonged delivery or maternal fatigue)	2 (4.4%)
Elective Caesarean section	9 (20.0%)
Neonates	
Gestation (weeks)	39.0 (36.3-41.4)
Birth weight (g)	2770 (2006-3695)
Small for gestational age	6 (13.3%)
Male	26 (57.8%)
One-minute Apgar score	8 (7-10)
Five-minute Apgar score	9.5 (8-10)
Umbilical artery pH	7.29 (7.095-7.437)
Use of oxygen within five minutes of birth	3 (6.7%)

Data are presented as mean  $\pm$  standard deviation, median (range) or number (frequency).

completed and recorded in the Japan Obstetric Compensation System for Cerebral Palsy by the end of 2016. We focused on infants who were apparently healthy at birth, the criteria for this were that their 5-minute Apgar scores were seven or more, with normal umbilical cord blood gases and they did not require neonatal resuscitation up to five minutes after birth.

By the end of 2016, case analysis reports on 1191 infants had been published (Figure 1), but we excluded nine infants born in

birthing centres, because they provide quite different medical care to hospitals and clinics and the number was quite small. In Japan, about half of the babies are born in hospitals and half are born in clinics, which are generally small-scale private facilities that specialise in obstetrics. About 1% are born in birthing centres. Of the remaining 1182 infants, 326 met our definition of being healthy at birth. However, 75 required neonatal resuscitation because of a postnatal collapse more than five minutes after birth. We excluded five infants who required neonatal intensive care unit admission because of low birth weight, multiple birth or mild morphological abnormalities. We also excluded 16 infants who presented with metabolic acidosis, characterised by an umbilical cord blood pH of less than 7.0 and, or, a base deficit that was higher than 16 mmol/L or mild respiratory impairment at birth. In addition, nine infants who underwent resuscitation after discharge from delivery facilities were excluded. The final number of apparently healthy infants who developed severe cerebral palsy due to SUPC was 45, and these accounted for 3.8% of the 1182 certified cerebral palsy cases. We retrospectively analysed the clinical manifestations of these 45 subjects based on the database.

In this study, cerebral palsy referred to a permanent and variable disorder of motor function or posture of the infant caused by a non-progressive cerebral lesion that developed during conception or the neonatal period. Patients with motor retardation that was progressive, transient or normalised in the future were not included.

Sudden unexpected postnatal collapse was confirmed when any term or near-term infant, who was deemed healthy at birth with a normal 5-minute Apgar score, collapsed unexpectedly within the first 7 days of life. These patients either died or went on to require intensive care or develop encephalopathy.<sup>5</sup> SUPC included both severe apparent life-threatening events and sudden unexpected deaths in infancy that occurred within the first postnatal week.<sup>6</sup>

Early skin-to-skin contact (SSC) was defined as SCC between mothers and their infants up to 24 hours after birth. Neonates who were naked, with or without a diaper, were placed prone on their mother's bare chest between her breasts, and the infant was kept dry and warm before and during SSC.<sup>7</sup>

Neonatal resuscitation was defined as any artificial ventilation, including chest compression, tracheal intubation and adrenaline infusion, which was performed in infants with apnoea, respiratory failure, bradycardia or cardiopulmonary arrest.

## 2.2 | Ethics

This study was approved by the institutional review board of the Japan Council for Quality Health Care. Written informed consent was not required from the parents as the data source was anonymised. In addition, the information on this research was posted on the website of the Japan Obstetric Compensation System for Cerebral Palsy and the parents were guaranteed the opportunity to refuse participation by using an opt-out system.

### 3 | RESULTS

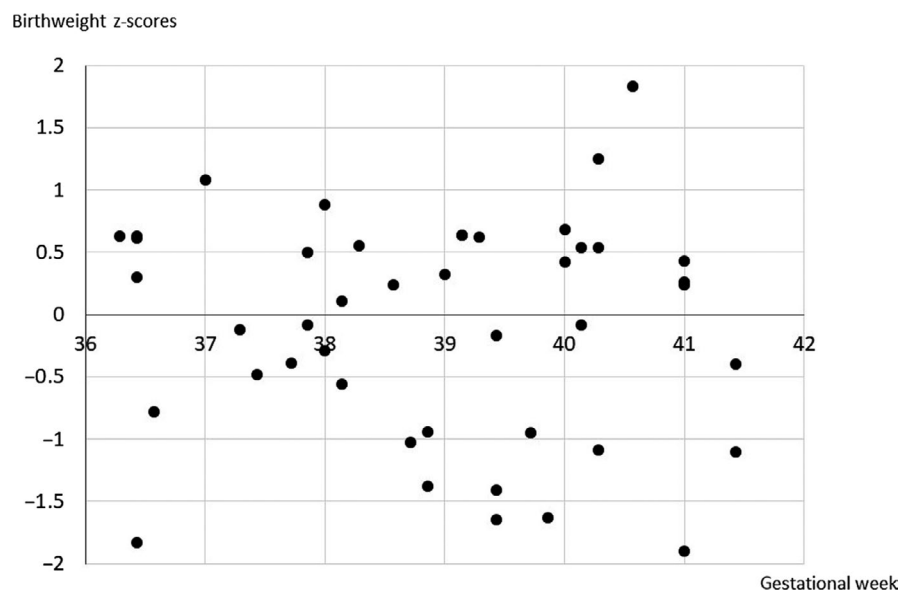
The median age of the 45 mothers was 32 years (range 20-45), and 33 were primiparas (Table 1). Abnormalities of foetal heart rhythm during labour were observed in 27 cases, and vacuum delivery was performed in four cases and emergency Caesarean sections were performed in another four cases (Table 1). The distribution of gestational ages and birthweight standard deviation (z-scores) are shown in Figure 2.

Resuscitation was initiated within two hours of birth in 11 (24.4%) infants and within 24 hours of birth in 31 (68.9%) infants (Figure 3). Of the 45 infants who collapsed, 27 developed SUPC during rooming-in with their mothers and 10 of these developed it during early SSC (Table 2). At the time of collapse, 15 infants

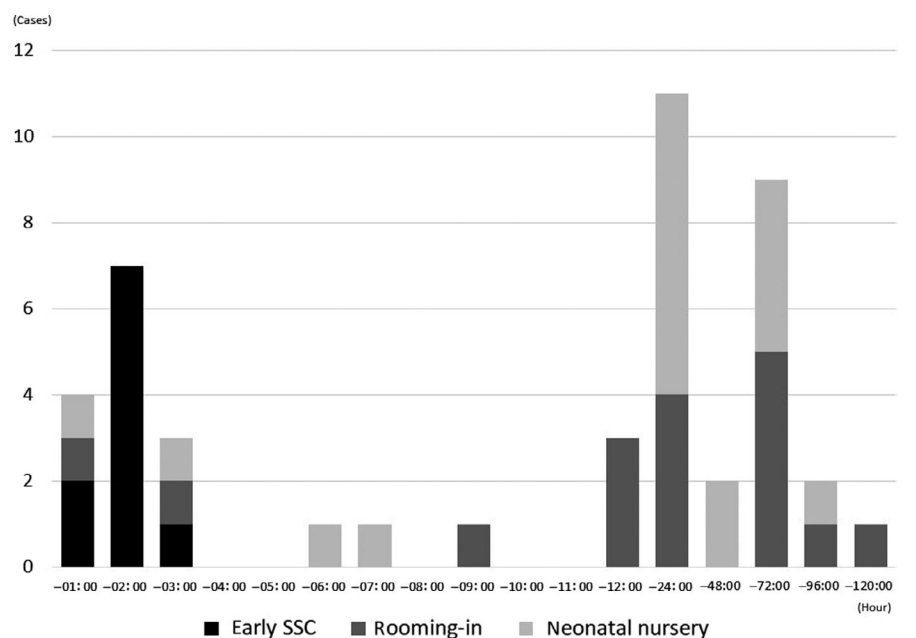
were being electronically monitored (33.3%). These included five infants who were managed separately from their mothers in the neonatal nursery and were receiving electronic apnoea monitoring as part of the nursery's routine management procedures. A pulse oximeter was used for the other 10 infants, because of minor symptoms such as hypoglycaemia, fever, cyanotic episodes or lethargy that appeared during rooming-in management. None of the infants who collapsed during early SSC or co-bedding were being monitored at the time of their collapse. Only 10 of the 45 mothers noticed their infants' abnormal presentation, and the other 35 were identified by medical personnel (Table 2).

Table 3 shows the possible causes of SUPC and cerebral palsy confirmed by the Committee in 11 of the 45 cases. The causes were not identified in the remaining 34 cases.

**FIGURE 2** The distribution of gestational age and birthweight z-scores



**FIGURE 3** The distribution of the timing of the initiation of neonatal resuscitation



**TABLE 2** Circumstances at the onset of sudden unexpected collapse of infants

Variables	Cases (n = 45)
The timing of initiation of neonatal resuscitation	
Within two hours	11 (24.4%)
Within three hours	14 (31.1%)
Within 24 hours	31 (68.9%)
Circumstances at the onset of collapse	
Rooming-in management	27 (60.0%)
Early skin-to-skin contact	10 (22.2%)
Co-bedding	11 (24.4%)
Holding on mother's breast/abdomen	2 (4.4%)
Supine position in baby cot	1 (2.2%)
Unknown	3 (6.7%)
Neonatal nursery	18 (40.0%)
Prone position	2 (4.4%)
Supine position	1 (2.2%)
Unknown	15 (33.3%)
Electronic monitoring	15 (33.3%)
Discovered by medical professionals	35 (77.8%)

Data are presented as number (frequency).

Of the 10 infants who developed SUPC during early SSC, eight developed SUPC during the night shift or in the morning between 5 PM and 9 AM (Table 4). When they were found to be in poor condition, nine of the 10 infants were being breastfed directly by their mothers and one was vomiting amniotic fluid. In all 10 cases, the medical personnel were not in attendance during early SSC, meaning that mothers and infants were left alone in the delivery room when the collapse occurred. Moreover, only two mothers noticed their infants' abnormal condition and the other eight mothers were unaware of any problems until medical personnel alerted them. Of the 10 mothers, nine were primiparous. Four mothers claimed intense fatigue, and one mother was sleeping at the time of SUPC. Only two mothers had been educated in advance about the warning signs during early SSC. One mother reported insufficient advice, and one reported not receiving advice. The remaining six mothers could not remember if they had received advice in advance.

## 4 | DISCUSSION

This study used data from The Japan Obstetric Compensation System for Cerebral Palsy from its inception in 2009 to the end of 2016. We identified 45 severe cerebral palsy cases due to SUPC, even though the patients appeared to be healthy at birth. This showed that SUPC accounted for 3.8% of severe cerebral palsy covered by the compensation system in Japan. It should be pointed out that that the system only provides no-fault compensation for children with severe disabilities due to cerebral palsy as a

**TABLE 3** The causes of cerebral palsy

Variables	Cases (n = 45)
The cause of cerebral palsy	
Single factor	
Hyperpotassaemia	3 (6.7%)
Group B streptococcus infections	2 (4.4%)
Umbilical cord abnormalities	1 (2.2%)
Persistent pulmonary hypertension of neonate	1 (2.2%)
Cardiovascular failure due to ventricular tachycardia	1 (2.2%)
Foeto-maternal blood transfusion	1 (2.2%)
Placental dysfunction	1 (2.2%)
Multifactorial	
Hyperinsulinaemia and cardiac arrest	1 (2.2%)
Unexplained	34 (75.6%)

Data are presented as number (frequency).

result of trauma during labour and delivery. The incidence of SUPC among healthy infants has not been investigated in depth in Japan. However, the incidence of SUPC in newborn infants has been reported for each 100 000 births in a number of other countries: 3.2 within two hours of birth in France,<sup>8</sup> 2 within 12 hours of birth in the UK and Ireland,<sup>9</sup> 6 within 24 hours of birth in Germany<sup>10</sup> and 21 within 24 hours of birth in Stockholm, Sweden.<sup>11</sup> A review article by Herlenius and Kuhn, that comprised 26 papers covering 398, showed that the incidence of SUPC ranged from 2.6 to 133 cases per 100 000 births.<sup>6</sup> We assume that this large difference was due to variations in the backgrounds of the study populations and the definitions of SUPC that were used. For example, some studies used the five-minute Apgar score and others use the 10-minute Apgar score in their SUPC definitions. According to the policy statement from the American Academy of Pediatrics, a 5-minute Apgar score of seven or higher is considered normal, even though this cannot be used to establish a diagnosis of asphyxia.<sup>12</sup> For this reason, we defined apparently healthy infants as those with normal five-minute Apgar scores and normal umbilical cord blood gases. This was because 10-minute Apgar scores are rarely recorded for healthy newborn infants in most Japanese delivery facilities.

Another paper<sup>13</sup> on the German survey by Poets et al, referenced above,<sup>10</sup> reported that more than half of SUPC occurred within two hours of birth. In our study, the onset of SUPC was concentrated in the first three hours of birth, mostly during early SSC.

A meta-analysis published by the Cochrane Systematic Review<sup>7</sup> clearly demonstrated the effect of early SSC on increased breastfeeding duration and maternal and infant bonding. A number of delivery facilities in Japan have introduced early SSC and rooming-in immediately after delivery.

Of the 10 infants with SUPC during early SSC, nine were being breastfed by their mothers and one was vomiting amniotic fluid when they were found in a poor condition. Even though the causes

**TABLE 4** Summary of characteristics of infants who developed sudden unexpected collapse during early skin-to-skin contact with their mothers

Case	1	2	3	4	5	6	7	8	9	10
Time after birth when infant was last seen healthy	34 min	45 min	54 min	50 min	67 min	84 min	72 min	92 min	84 min	94 min
Time after birth when infant collapsed	49 min	55 min	64 min	75 min	92 min	101 min	104 min	107 min	114 min	117 min
Time between last seeing healthy infant and collapse	15 min	10 min	10 min	25 min	25 min	17 min	32 min	15 min	30 min	23 min
Shift pattern when infant collapsed	Weekday	Weekday	Holiday	Weekday	Holiday	Weekday	Weekday	Weekday	Holiday	Weekday
Delivery facility	Night shift	Day shift	Night shift	Night shift	Night shift	Night shift	Night shift	Day shift	Night shift	Night shift
First person to notice collapse	Hospital	Hospital	Clinic	Hospital	Hospital	Hospital	Hospital	Hospital	Hospital	Clinic
Other family members than mother in attendance when infant collapsed	Mother	Nurse	Midwife	Midwife	Midwife	Midwife	Mother	Midwife	Midwife	Nurse
Alert state of mother	No	Husband	Husband	Unknown	Unknown	Unknown	Unknown	No	Husband	No
Prior guidance on risks of early skin-to-skin contact	Fatigue	Awake	Fatigue	Awake	Awake	Awake	Awake	Awake	Fatigue	Sleeping
Breastfeeding at the onset of collapse	Unknown	No	Yes	Unknown	Unknown	No	Unknown	Yes	Unknown	Unknown
Vomiting at the onset of collapse	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Maternal age (years)	Yes	Yes	No	No	No	No	No	No	No	No
Primipara	24	30	27	40	32	27	32	32	25	27
Maternal body mass index (%)	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Painless delivery	19.7	25.0	23.6	17.2	21.2	19.5	21.7	21.4	18.4	19.1
Induced delivery	No	No	No	No	No	No	No	Yes	No	No
Abnormalities of foetal heart rate	No	No	No	Yes	Yes	Yes	Yes	Yes	No	Yes
Length of delivery	Yes	Yes	No	Yes	Yes	No	No	Yes	No	No
Gestational age at birth, weeks and days	9 h 5 min	7 h 3 min	5 h 20 min	2 h 58 min	8 h 47 min	6 h 46 min	27 h 38 min	2 h 51 min	8 h 45 min	20 h 38 min
Birth weight, grams	38 + 0	38 + 5	37 + 2	40 + 2	38 + 4	40 + 1	40 w 4 d	39 w 1 d	37 w 6 d	39 w 3 d
Gender	3092 g	2685 g	2552 g	2770 g	2884 g	3080 g	3695 g	3092 g	2855 g	2895 g
Neonatal transfer	Male	Male	Female	Male	Female	Male	Female	Female	Female	Female
	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes



of SUPC were not determined in these cases, we assume that they may have been associated with airway obstruction, due to an inappropriate breastfeeding technique and an asphyxiating position during early SSC.

Interestingly, nine out of the 10 mothers were primiparous and this 90% was substantially higher than the proportion of primiparous mothers in Japan, which is 50%. Primiparous mothers lack the experience and knowledge to avoid airway obstruction while they are feeding and holding their baby, and extensive fatigue after a long delivery times has been associated with SUPC in infants. The period soon after birth is the most unstable period for newborn infants because they have to adapt to *ex-utero* respiratory and circulatory functioning. During early SSC, mothers and infants should not be left alone in the delivery room and close observation of the consciousness of the mother and the condition of the infant is essential.

In this case series, electronic monitoring was being performed on 15 infants when they collapsed. They included five infants who were managed separately from their mother in the neonatal nursery and were on electronic apnoea monitors from the time of birth. In some facilities in Japan, electronic apnoea monitors are routinely used for all newborn infants, especially when they are managed separately from their mothers and that was the policy in this study facility. The other 10 infants who received electronic monitoring were on a pulse oximeter because of some minor abnormalities before they collapsed. The fact that these 15 infants subsequently developed severe cerebral palsy suggests that electronic monitors may be useful for detecting changes in respiratory and circulatory dynamics in infants, but they cannot prevent SUPC and cerebral palsy. Because SUPC occurred in 10 infants who were on pulse oximeters because of abnormal symptoms before they collapsed, they may not have been as healthy as they initially appeared to be.

The Japan Society of Perinatal and Neonatal Medicine published a handbook called *Points to Bear in Mind in Regard to the Implementation of Early Mother-Infant Skin-to-Skin Contact* in 2012.<sup>14</sup> The handbook provides the following advice on detecting abnormal signs in infants at an early stage. First, infants should be carefully observed with electronic monitoring or by medical professionals in the delivery room during early SSC. Medical professionals should not entrust the observation of infants to just the mother. Second, sufficient guidance should be provided to mothers and families on the risk of early SSC as well as the effectiveness. They should also be advised on the correct procedure for breastfeeding and positioning. Thirdly, all medical staff working in delivery facilities must be educated on neonatal resuscitation and each facility should provide a manual on this based that is based on their hospital and its systems.

There were several limitations to this study. Even though the no-fault compensation system covers 99.9% of the delivery facilities in Japan, the number of deliveries in each participating facility was not reported. Some applicants who applied for compensation from 2009 to 2014 were still under review at the end of 2016, and the total number of cases is likely to increase. Therefore, we

could not estimate the incidence of severe cerebral palsy due to SUPC. Furthermore, SUPC may cause minor disabilities that are not as serious as severe cerebral palsy and none of these cases were included in the present study. As a result, the incidence of SUPC in the Japanese population cannot be estimated from the current study, as it only focused on severe cerebral palsy due to SUPC. Another major limitation was that we could not conduct a case-control study, because no data were available on the infants who did not develop cerebral palsy. This meant that we could not analyse the causal factors of SUPC and cerebral palsy in infants who were apparently healthy at birth. Furthermore, detailed information on each subject was insufficient, because the data were not based on case analysis reports for the purposes of compensation and not full medical reports. Finally, the number of the subjects was small.

Despite those limitations, we believe that this was the first report to focus on apparently healthy infants who subsequently developed severe cerebral palsy from a large-scale national dataset in Japan. The information from this case series has strong implications for how facilities should manage infants and mothers after birth.

## 5 | CONCLUSION

This national study of children with cerebral palsy who appeared healthy at birth found that unsupervised breastfeeding was a common factor in cases of SUPC during early SSC. The efficacy of early SSC for promoting breastfeeding and maternal and infant bonding has been scientifically proven. It is not our intention to use our study results to undermine the efficacy of early SSC or prevent its implementation, as it is a legitimate right for both infants and mothers. However, our findings do underline the importance of careful observation during early SSC as the general condition of newborn infants can change rapidly and dramatically, even when they appear healthy just after delivery. All medical staff working in delivery facilities need to be aware of the links between early SSC, SUPC and the subsequent development of cerebral palsy.

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## CONFLICTS OF INTEREST

The authors have no conflicts of interest to declare.

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