

# Oropharyngeal Dysphagia in Breastfeeding Neonates with Hypoxic-Ischemic Encephalopathy on Therapeutic Hypothermia

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## Abstract

**Purpose:** To identify symptoms of oropharyngeal dysphagia (OPD) in breastfeeding neonates with hypoxic-ischemic encephalopathy (HIE) on therapeutic hypothermia (TH). Early identification of feeding problems in neonates with HIE by speech-language therapists (SLTs) may prevent secondary complications of OPD such as aspiration pneumonia and death.

**Materials and Methods:** Twenty-eight full-term neonates with HIE (mean chronological age = 4.5 days) and 30 healthy term controls were prospectively recruited for this case-control study. Participants with HIE (mild [ $n = 15$ ], moderate [ $n = 11$ ], severe [ $n = 2$ ]), diagnosed by pediatricians, received whole-body TH. Feeding was clinically evaluated by an SLT using the Preterm Infant Breastfeeding Behavior Scale.

**Results:** Twenty-five neonates (89.2%) had at least one symptom of OPD. Falling asleep during feeding, noticeable oral secretions, coughing, and flaring nostrils were symptoms of OPD most frequently identified. The HIE group displayed limited arousal during breastfeeding and had less obvious rooting, shallower latching onto the breast, and more single sucks in comparison to term newborns. The HIE group had significantly more closed eyes and minimal movement during breastfeeding, while controls showed the quiet-alert state ideal for breastfeeding.

**Conclusions:** OPD was identified in the majority of infants with HIE. Underlying the appearance of an inactive neonate with HIE may be OPD that could be overlooked if not investigated. Interprofessional collaboration between SLTs, pediatricians and nurses to determine feeding-readiness is imperative. SLTs may assist in decision-making to improve safety of breastfeeding in this population. This study contributes to the small body of research on early breastfeeding of neonates with HIE.

**Keywords:** hypoxic ischemic encephalopathy, therapeutic hypothermia, breastfeeding, neonate, symptoms of oropharyngeal dysphagia

## Introduction

THERAPEUTIC HYPOTHERMIA (TH) is regarded as standard care for infants with moderate to severe hypoxic-ischemic encephalopathy (HIE), even in resource-limited settings.<sup>1-5</sup> Feeding difficulties of infants with HIE not only poses a risk for gastrointestinal complications, but also represents a possible neurological cause of oropharyngeal dysphagia (OPD).<sup>5-8</sup> There are variations in feeding practices during TH for infants with HIE.<sup>5</sup> However, literature on early feeding characteristics and the introduction of oral feeding,

particularly the symptoms of OPD during breastfeeding, in neonates with HIE during TH, is scarce.

Disturbances of sucking, swallowing and tongue movements in neonates with HIE are probably related to brainstem involvement.<sup>9,10</sup> It appears that maturation of normal swallowing is also affected in full-term infants with HIE.<sup>11</sup> Early oral feeding may therefore be compromised in neonates with HIE and the presence of swallowing and feeding difficulties, or OPD should be investigated.

Available literature is limited to infants with nonspecific neurological disorders and little is specified about neonates

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with HIE. Infants with injuries of the basal ganglia, pons, and thalamus are at risk of ongoing OPD, with a 91% probability of long-term tube feeding.<sup>11</sup> Newborns with neurological problems may have sucking difficulties, absent swallowing, and an absent reflexive cough.<sup>7</sup> Symptoms such as pooling, or anterior oral spillage of liquid, prolonged feeding times, coughing, gagging, emesis, reflux, nasal regurgitation, apnea or respiratory problems during feeding, straining during feeding, and inadequate weight gain may be observed in infants with neurological disturbances.<sup>7,9,11–13</sup> In addition, infants with neurological disorders may have poor endurance, leading to fatigue, and consequently increasing their risk of aspiration.<sup>12</sup> The specific oral feeding skills of neonates with HIE are not clearly described.

Furthermore, some controversy in literature exists as the ideal feeding regime for infants with HIE on TH has not been established yet.<sup>14,15</sup> Total parenteral nutrition is often preferred.<sup>3,14</sup> While enteral nutrition (EN) is shown to be safe and feasible during TH in one study, another recommends no EN until after rewarming.<sup>5,14</sup> TH has been found to improve the feeding tolerance of infants with moderate to severe HIE.<sup>16</sup> Perinatal complications such as asphyxiation, or HIE, can affect breastfeeding initiation.<sup>17</sup>

A case study in South Africa found that a combination of EN and breastfeeding was used for a neonate with HIE who received TH.<sup>18</sup> Further review of literature revealed limited information on early oral feeding attempts, and more specifically the breastfeeding abilities in neonates with HIE. Research on oral feeding in neonatal care is often limited to studies using bottle feeding and findings are thus not generalizable to breastfeeding.<sup>19</sup>

A need exists for a population-based study, describing early feeding characteristics of neonates with HIE during TH, as well as possible symptoms of OPD.<sup>20</sup>

Krüger et al.,<sup>18</sup> reported early breastfeeding difficulties in a single infant with mild HIE who underwent TH. Despite having mild HIE, the infant still presented with oral stage dysphagia and breastfeeding difficulty, leading to longer hospitalization.<sup>18</sup> Early feeding difficulties following HIE is associated with poor outcome.<sup>3</sup> There is a high incidence of HIE in low- and middle-income countries, and early intervention for feeding problems by the speech-language therapist (SLT) is important. Early identification may prevent secondary complications of OPD such as aspiration pneumonia and death. The aim of the present study was to identify directly observable symptoms of OPD in breastfeeding neonates with HIE on TH.

## Materials and Methods

### Setting

Data were collected prospectively in neonatal nurseries and postnatal wards of two academic hospitals in Pretoria, South Africa. These baby-friendly hospitals encourage exclusive breastfeeding. Neonates diagnosed with HIE, and deemed suitable candidates by the managing pediatrician/neonatologist according to the hospital protocol for TH, receive whole body TH for 72 hours. Currently, there are no locally published guidelines for the implementation of TH in a resource-constrained South African context.<sup>21</sup>

Local eligibility criteria for TH are based on the six largest published trials to date and include the following: (1) infants

with a gestation of  $\geq 36$  weeks, aged  $< 6$  hours; (2) who have signs of possible intrapartum hypoxia-ischemia (Apgar of  $\leq 5$  at 10 minutes, or who require continued resuscitation at 10 minutes, or who have severe acidosis [pH  $< 7$ /base deficit  $\geq 16$ ]); and (3) who have an abnormal neurological assessment (moderate and/or severe encephalopathy, or clinical seizures, or an abnormal amplitude-integrated electroencephalography).<sup>21</sup>

### Participants

A sample of 58 full-term neonates ( $\geq 37$  weeks)<sup>22</sup> were recruited using nonprobability purposive sampling for this case-control study.<sup>23</sup> Twenty-eight infants with HIE were included in the research group (RG). Thirty term, healthy newborns were recruited as a control group (CG). The groups did not differ significantly in mean birth weight, gestational age, or chronological age (Table 1), which ensured differences in maturation did not influence findings.

A pediatrician independently diagnosed HIE using criteria by Martinez-Biarge et al.<sup>24</sup> and the HIE score.<sup>25</sup> All RG participants received whole-body TH. There is currently no consensus for the ideal numerical value for eligibility for TH when using the HIE score by Thompson et al.<sup>25</sup> When using the HIE score<sup>25</sup> to determine eligibility for TH, tertiary institutions with more resources such as the setting in the present study, may choose a lower threshold.<sup>21</sup> Neonates with meningitis, and metabolic, or congenital disorders were excluded. Neonates with HIV-exposure were included as no evidence could be found that HIV-exposure influences feeding in the neonatal period.

More than half of both groups were male. Most of the RG had 72 hours of TH ( $n=24$ ; 85.72%). Infants who did not receive 72 hours of TH were those who became unstable and a pediatrician/neonatologist decided to discontinue the treatment. More than half of the RG had mild HIE ( $n=15$ ; 53.57%). There was no significant difference between the two groups based on mode of delivery ( $p=0.0796$ ). EN was introduced to 17 of the RG participants (60.7%) during TH recommended by the pediatrician/neonatologist in charge and did not form part of a standardized protocol for the hospital.

### Data-collection

Voluntary participation was requested from neonates' parents. Institutional ethical clearance was obtained, and data were collected prospectively by the same SLT for both groups.<sup>18</sup> When deemed ready to feed orally by a pediatrician/neonatologist, a clinical feeding evaluation was conducted either during or following TH. Some of the RG participants were first orally fed during TH and others were only introduced to oral feeding following rewarming (mean chronological age of RG = 4.5 days [SD = 2.60]).

The Preterm Infant Breastfeeding Behavior Scale (PIBBS)<sup>27</sup> was used to document observable breastfeeding skills. It is a comprehensive tool for use by professionals and mothers and provides detailed information on the oral motor competence of infants during breastfeeding.<sup>19</sup> The PIBBS<sup>27</sup> has good reliability and validity and is suitable to use with preterm as well as full-term neonates.<sup>19</sup> Participants were observed before the feeding session. Thereafter, observations of breastfeeding were conducted using guidelines by Nyqvist et al.<sup>27</sup> for both the RG and CG. The SLT was positioned close

TABLE 1. PARTICIPANT CHARACTERISTICS (n=58)

	RG (n=28)	CG (n=30)	p
Mode, n (%)			0.0796
Normal vaginal	8 (28.57)	14 (46.67)	
Assisted	9 (32.14)	2 (6.66)	
Caesarean	11 (39.29)	14 (46.67)	
Male, n (%)	16 (57.14)	17 (56.66)	-
Mean birth weight (kg), mean (SD)	3.42 (0.45)	3.39 (0.46)	0.792
Mean gestation (weeks), mean (SD)	40.3 (1.67)	40.3 (1.67)	0.251
Mean chronological age (days), mean (SD)	4.5 (2.60)	3.4 (0.97)	0.057
HIE stage, <sup>26</sup> n (%)		—	—
Mild	15 (53.57)		
Moderate	11 (39.29)		
Severe	2 (7.14)		
TH duration (hours), n (%)		—	—
4	1 (3.57)		
36	1 (3.57)		
48	2 (7.14)		
72	24 (85.72)		
Maternal age (years), mean (SD)	26.8 (6.25)	28.0 (5.61)	0.453
HIV-exposure, n (%)	8 (28.57)	9 (30.00)	—

CG, control group; HIE, hypoxic-ischemic encephalopathy; RG, research group; SD, standard deviation; TH, therapeutic hypothermia.

enough to observe the neonate's mouth and face during breastfeeding, but stood or sat in a way to not disturb feeding. The mother was also asked whether she is able to perceive the letdown reflex during her feeding experience with the infant.

Parents were briefly interviewed and hospital files accessed for relevant demographic information,<sup>18</sup> and the HIE score,<sup>25</sup> calculated independently by the pediatrician/neonatologist in the case of the RG. Infants identified with OPD were referred for intervention.

#### Data-analysis

The *t*-test, Fisher's exact test, and Wilcoxon signed rank test were used to determine differences between groups on the PIBBS using SAS<sup>®</sup> version 9.3 (SAS Institute, Inc., Cary, NC). *p*-values of  $\leq 0.05$  were deemed significant. Descriptive statistics were also used.

#### Results

##### Observations of breastfeeding of neonates with HIE

All RG participants received EN at each feed using expressed breast milk (EBM) after direct breastfeeding was attempted. Participants also received EBM via cup feeding to supplement direct breastfeeding. Mothers lodge in the hospital and therefore EBM was available. Most RG participants ( $n=27$ ; 96.4%) were positioned using the cradle hold. Feeding occurred 3 hourly according to the neonatal nursery schedule.

The majority of infants with HIE presented with limited arousal ( $n=20$ ; 71.42%) and infrequent irritability ( $n=18$ ; 64.28%) as reported by mothers (Table 2). Most displayed hypertonicity ( $n=22$ ; 78.5%). More than half ( $n=15$ ; 53.57%) had noticeable oral secretions indicating decreased swallowing. Coughing ( $n=7$ ; 25.0%) and flaring nostrils ( $n=7$ ; 25.0%) were the most frequently observed symptoms of OPD. Comparative results provide stronger evidence for

swallowing difficulties as is illustrated by the following findings.

##### Comparison of breastfeeding skills between groups

The comparison revealed significant differences between the two groups on almost all aspects of the PIBBS.

The RG showed significantly less obvious rooting to the breast ( $p<0.0001$ ) (Table 3). The RG also showed significantly less of the ideal deep latch onto the whole nipple

TABLE 2. OBSERVATIONS OF NEONATES WITH HIE DURING BREASTFEEDING (n=28)

Characteristic	n (%)
Arousal	
Falls asleep, 4–5 minutes	20 (71.42)
Sustained, 10 minutes	5 (17.85)
Fluctuating/intermittent	3 (10.71)
Irritability	
Mostly quiet	5 (17.85)
Infrequent, calms easily	18 (64.28)
Frequent, calms with holding	5 (17.85)
Muscle tone	
Hyper	22 (78.5)
Hypo	3 (10.71)
Normal	5 (17.85)
Noticeable oral secretions	15 (53.57)
Coughing	7 (25.0)
Gagging/emesis	0 (0.0)
Noisy breathing	2 (7.14)
Gurgly voice	1 (3.57)
Flaring nostrils	7 (25.0)
Fussing behavior	6 (21.4)
Feeding duration (minutes)	
0–20	10 (35.71)
30–40	13 (46.42)
<45	5 (17.85)

TABLE 3. COMPARISON OF BREASTFEEDING SKILLS USING THE PIBBS ( $n=58$ )

Breastfeeding skills, n (%)	RG (n=28)	CG (n=30)	p
Rooting			
None	9 (32.14)	0 (0.00)	0.0006 <sup>a</sup>
Some	14 (50.00)	8 (26.67)	0.1040
Obvious	5 (17.86)	22 (73.33)	<0.0001 <sup>a</sup>
Latching depth			
None	4 (14.29)	0 (0.00)	0.0483 <sup>a</sup>
Part nipple	7 (25.00)	0 (0.00)	0.0039 <sup>a</sup>
Whole nipple	10 (35.71)	6 (20.00)	0.2432
Some areola	7 (25.00)	24 (80.00)	<0.0001 <sup>a</sup>
Latching duration (minutes)			
None	7 (25.00)	0 (0.00)	0.0039 <sup>a</sup>
<1	16 (57.84)	30 (100.00)	<0.001 <sup>a</sup>
Sucking			
None	6 (21.43)	0 (0.00)	0.0093 <sup>a</sup>
Licking	2 (7.14)	0 (0.00)	0.2287
Single sucks	10 (35.71)	0 (0.00)	0.0003 <sup>a</sup>
Short bursts	6 (21.43)	13 (43.33)	0.0973
Long bursts	4 (14.29)	17 (56.67)	0.0010 <sup>a</sup>
Mean longest sucking burst (SD) [number of sucks]	7.82 (7.62)	16.73 (7.42)	<0.0001 <sup>a</sup>
Swallowing			
None	8 (28.57)	0 (0.00)	0.0016 <sup>a</sup>
Occasional	11 (39.29)	0 (0.00)	<0.0001 <sup>a</sup>
Repeated	9 (32.14)	30 (100.00)	<0.0001 <sup>a</sup>
Letdown reflex experienced by mother	6 (21.43)	26 (86.67)	<0.001 <sup>a</sup>

<sup>a</sup>Statistically significant.

and some of the areola ( $p<0.0001$ ) than the CG. The entire CG latched onto the breast for longer than 1 minute, while only 57.84% of the RG performed similarly ( $p<0.0001$ ). The RG used significantly more single sucks ( $p=0.0003$ ) and short sucking bursts rather than the ideal long sucking bursts ( $p=0.0010$ ) mostly demonstrated by the controls. The majority of the RG showed occasional ( $n=11$ ; 39.29%) or no swallowing ( $n=8$ ; 28.57%). The RG therefore displayed symptoms of OPD during breastfeeding while the CG showed no feeding difficulty. Differences in the groups' general behavior during breastfeeding are displayed in Table 4.

The RG had significantly more infants with closed eyes and minimal movement during breastfeeding ( $p<0.0009$ ), while the controls showed significantly more ideal feeding behavior of open eyes, eye contact with minimal movement ( $p<0.0001$ ) (Table 4). Minimal movement was observed in both groups. The control group showed minimal movements with open eyes characteristic of the quiet-alert state, which is the ideal behavior for oral feeding, while the RG displayed mostly minimal movement and closed eyes related to quiet sleep.

## Discussion

The study not only described early breastfeeding of infants with HIE on TH but also identified at least one symptom of OPD in 25 of 28 neonates (89.2%) in the sample. Most infants were diagnosed with mild ( $n=15$ ) and moderate ( $n=11$ ) HIE, and only two (7.14%) had severe HIE. The symptoms of OPD included falling asleep during feeding, the presence of visible oral secretions, coughing during feeding, flaring nostrils, and the absence of frequent swallowing during breastfeeding.

The majority of the RG showed either only occasional swallowing or no swallowing, which is concerning as they were medically deemed ready to commence oral feeding. These findings concur with available publications on a similar population.<sup>11,18</sup> More than half of the RG ( $n=15$ ; 53.57%) had noticeable oral secretions and a quarter ( $n=7$ ; 25%) showed coughing. Coughing and choking may point to aspiration during feeding.<sup>28</sup> Poor control of oral secretions may be a symptom of swallowing dysfunction, inadequate oral-motor control, and is indicative of unsafe oral feeding.<sup>28</sup> The RG also used significantly more single sucks ( $p=0.0003$ )

TABLE 4. GENERAL BEHAVIOR DURING BREASTFEEDING USING PIBBS ( $n=58$ )

General behavior, n (%)	RG (n=28)	CG (n=30)	p
Closed eyes, no movement	11 (39.29)	1 (3.33)	0.0009 <sup>a</sup>
Drowsy, minimal movement	12 (42.86)	13 (43.33)	1.000
Open eyes, dull/glazed look, minimal movement	3 (10.71)	0 (0.00)	0.1062
Wide open eyes, eye contact, minimal movement	1 (3.57)	16 (53.33)	<0.0001 <sup>a</sup>
Closed eyes, active movement	1 (3.57)	0 (0.00)	0.4828

<sup>a</sup>Statistically significant.

and short sucking bursts rather than the ideal long sucking bursts ( $p=0.0010$ ) mostly used by the CG during breastfeeding. Single sucks and short sucking bursts may also be linked to swallowing problems in infants.<sup>28</sup>

Apart from identifying symptoms of OPD, this study also found oral motor difficulties in the participants with HIE when comparing their breastfeeding attempts to typical healthy newborn infants' breastfeeding using the PIBBS. The RG showed significantly less obvious rooting to the breast ( $p<0.0001$ ) (Table 3). Rooting is important for successful latching during breastfeeding.<sup>28</sup> This finding may explain why the RG also had significantly less of the ideal deep latch onto the whole nipple and some of the areola ( $p<0.0001$ ) than the controls.

Problematic neural integration, as found in an infant with HIE, may explain the diminished rooting and opening of the mouth for breastfeeding observed in this group.<sup>28</sup> Most of the RG also showed hypertonicity (78.5%). Infants with central nervous system disorders and abnormal muscle tone are at risk of oral sensorimotor dysfunction,<sup>9</sup> which may further contribute to possible OPD. The importance of the presence of the SLT in the neonatal nursery to assist in decision-making about feeding-readiness, is highlighted by these findings, as assessment and treatment of OPD in neonates remain the responsibility of the SLT.<sup>29</sup>

Central to the feeding of the HIE group was their apparent inactivity and their behavioral state characterized by closed eyes, minimal movement, as well as signs of drowsiness and quiet sleep during breastfeeding. Most participants with HIE had limited arousal according to maternal report ( $n=20$ ; 71.42%), which agreed with the finding on the PIBBS (Table 4) that the RG had significantly more infants with closed eyes and minimal movement related to the drowsy and sleep states when compared to the CG ( $p<0.0009$ ). Falling asleep during feeding is regarded as an infant stress signal and may negatively influence breastfeeding, as quiet-alert is the ideal state for feeding.<sup>9,28</sup> This finding is similar to that of a previous study of an infant with mild HIE presenting with feeding difficulties related to inadequate behavioral state regulation.<sup>18</sup>

Participants with HIE may have shown inactivity during feeding due to falling asleep, which may be related to either neurological compromise, the treatment of TH, the possible sedatory influence of medication, or a combination of these factors. Further research is therefore required. Underlying the appearance of an inactive, lethargic neonate with HIE, may be symptoms of OPD that could easily be overlooked if not investigated in depth, as the CG also showed limited movement during breastfeeding. The findings indicate that introduction of oral feeding should be appropriately timed to ensure the safety and health of neonates with HIE. Early assessment and intervention for breastfeeding difficulties, and more specifically OPD in this population by the SLT, are therefore crucial.

All participants with HIE were exclusively receiving breast milk using direct breastfeeding with EN and cup feeding, which is encouraging, as it agrees with recommendations by the South African Department of Health recommendations.<sup>30</sup> The prospective data collection in this study, the rather large sample of infants with HIE, and the close matching of the groups are strong points.

A limitation of the study is that the research hospitals did not use a standardized protocol for feeding neonates during

TH, and made use of the managing pediatrician/neonatologist to decide feeding-readiness, possibly leading to bias. A further limitation is that this study only used clinical measures to investigate feeding and swallowing, while future research using instrumental assessment may be valuable. Future research may also aim to longitudinally track the feeding progression of infants with HIE. This may ensure well-timed maternal counseling and the introduction of appropriate interventions.

The risks associated with the introduction of oral feeding in this population, such as the use of medication during TH, and other maternal factors should also be investigated as it was a limitation in this study. This study contributes to the small body of research on early breastfeeding of neonates with HIE on TH.

The breastfeeding skills and symptoms of OPD during breastfeeding of neonates with HIE, namely inadequate behavioral state, poor rooting, inadequate latching and sucking, and unsafe swallowing were presented. The variety of symptoms characterizing the number of breastfeeding difficulties and the severity thereof are noteworthy findings, as the majority of the RG had only mild HIE, a group who may be overlooked by clinicians with large caseloads. The finding agrees with previous research concluding that mild HIE has not been well studied.<sup>31</sup>

Breastfeeding support is therefore required for all infants who have HIE, including those with mild HIE, to ensure optimal breastfeeding outcomes. This study reiterates the importance of interprofessional collaboration between the SLT, the neonatologist and neonatal nurse, when oral feeding readiness is determined for neonates with HIE.

## Conclusions

The breastfeeding skills of a group of neonates with HIE were described and OPD was identified although infants were medically deemed ready to commence with oral feeding. Interprofessional collaboration among SLTs, pediatricians, and nurses is imperative when determining oral feeding readiness in infants with HIE in the neonatal nursery. The SLT may assist in decision-making to not only improve the safety of early breastfeeding attempts of neonates with HIE but also to support optimal breastfeeding outcomes in this population.

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