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ORIGINAL ARTICLE

Risk of dysphagia in a population of infants born preterm: characteristic risk factors in a tertiary NICU

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Abstract
Objective: To examine the prevalence and characteristics of dysphagia and suck-swallow-breath incoordination as phenotypes of oral feeding difficulties.
Method: A cross-sectional study with secondary data collected consecutively over 2 years from October 2020 to October 2022 to measure the prevalence of swallowing and oral feeding difficulty in preterm infants using Flexible endoscopic evaluation of swallowing examination at the tertiary Integrated Dysphagia Clinic.
Results: The prevalence of swallowing disorders was 25 % and the prevalence of suck-swallow-breath incoordination was 62.5 %. The significant risk factor that may show a possible correlation with oral feeding difficulty was mature post-menstrual age (p = 0.006) and longer length of stay (p = 0.004). The dominant percentage of upper airway abnormality and disorder were retropalatal collapse (40 %), laryngomalacia (42.5 %), paradoxical vocal cord movement (12.5 %), and gastroesophageal reflux disease (60 %). The dominant characteristic of oral motor examination and flexible endoscopic evaluation of swallowing examination was inadequate non-nutritive sucking (45 %), inadequate postural tone (35 %), and inadequate nutritive sucking (65 %).
Conclusion: Dysphagia in preterm infants is mostly observed in those with mature post-menstrual age, longer length of stay, and the presence of gastroesophageal reflux disease with inadequate non-nutritive sucking and nutritive sucking abilities. Suck-swallow-breath incoordination is primarily observed in those with immature post-menstrual age, a higher prevalence of cardiopulmonary comorbidity, and a higher prevalence of upper airway pathologies (laryngomalacia, paradoxical vocal cord movement) with inadequate nutritive sucking ability.

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Introduction

Preterm infants have a higher risk of developing complex comorbidities related to upper airway patency problems. Incoordination of the suck–swallow–breath (SSB) mechanism will affect oral feeding and swallowing difficulties.[1,2] Feeding problems caused by SSB incoordination will affect upper airway patency during feeding. Desaturation will occur if the respiratory rate of the infant is not maintained at an appropriate level during feeding. Some infants present with a laryngeal adductor reflex (LAR) abnormality, especially infants with congenital neurologic disorders, which increase the risk of milk aspiration. Long-term effects include respiratory problems, issues with growth, and developmental problems.[3,4]

Several etiologies underlying swallowing disorders in premature infants include (1) neonatal asphyxia, (2) underdeveloped anatomy and physiology, (3) neural and endocrine disorders, and (4) inflammation (i.e., pulmonary inflammation, such as pneumonia, gastrointestinal inflammation, such as gastroesophageal reflux disease (GERD) and necrotizing enterocolitis, and mechanical inflammation due to long-term use of an endotracheal tube).[5,6] These factors increase the risk of multiple comorbidities in prematurity, which require several treatments to stabilize, such as intravenous medication, gavage feeding, and mechanical ventilation, which are commonly performed in neonatal intensive care units (NICU). Prolonged use of mechanical ventilation reportedly increases the risk of delayed development of oral motor function due to mechanical inflammation of the upper airway, which causes LAR abnormality and delayed oral motor maturation. This is due to lung inflammation induced by mechanical ventilation (i.e., bronchopulmonary dysplasia or BPD). Thus, approaches for early diagnosis of feeding and swallowing ability in preterm infants should be considered.[4,7]

The video-fluoroscopy swallowing study (VFSS) is widely used to diagnose feeding and swallowing disorders in children and infants, and it is currently regarded as the gold-standard assessment for dysphagia and feeding problems. Radiation with contrast use, a period-restricted assessment, cannot be performed in a bedside assessment, barium allergic, and hardly used during breastfeeding are disadvantages of VFSS. Finally, the inability to assess upper airway anatomical conditions is also a barrier to VFSS use in NICUs. Later, flexible endoscopic evaluation of swallowing (FEES) was introduced. Some benefits of using FEES include widespread accessibility, support for bedside assessment, general radiation safety, an assessment of the upper airway structure, breastfeeding support, and use as a follow-up examination. Recent studies found that FEES is highly useful for diagnosing penetration and aspiration in preterm infants in NICUs, with an aspiration finding accuracy of approximately 92 % compared with that of VFSS. Thus, this is a reason for using FEES as a diagnostic tool for feeding and swallowing problems in preterm infants.[8-10]

SSB incoordination is a part of an infant's abnormal self-regulatory and balancing system. This adaptive behavior occurs because of interaction with environmental changes after birth. A normal self-regulatory and balanced system will allow for demand-feeding behavior. This ability requires behavioral state organization, attention, a rhythmic SSB pattern, and cardiorespiratory regulation. The maturation of behavioral and attention states enables infants to show signs of hunger or feeding readiness and to remain alert while feeding. A rhythmic suck–swallow pattern requires the development of non-nutritive sucking (NNS) and nutritive sucking (NS) abilities, which begin at approximately 32–36 weeks of gestational age. The SSB cycle develops in fully mature infants after 37 weeks of gestational age.[11,12] Lau et al.[1,2] compared term infant sucking and swallowing ability to that of preterm infants. Preterm infants typically have immature cardiorespiratory regulation, resulting in apnea and bradycardia when regulating cardiorespiratory effort during feeding. Because of SSB incoordination, this condition will cause oral feeding difficulties.[1,2,12]

SSB incoordination is a distinct form of dysphagia in prematurity, which sometimes clinically results in similar symptoms. The two phenotypes are identified as the etiology of preterm infant feeding difficulties. In this study, the authors examined the prevalence and characteristics of dysphagia and SSB incoordination as phenotypes of oral feeding difficulties, which have not been previously studied.

Methods

This was a cross-sectional study with secondary data collected consecutively over 2 years from October 2020 to October 2022. The study was conducted to measure the prevalence of swallowing and oral feeding difficulty in preterm infants using FEES examination at the tertiary Integrated Dysphagia Clinic, which included an ORL-HNS doctor, a rehabilitation doctor, an intensivist, and a neonatology doctor as the team for developmental care in the NICU to diagnose and manage feeding difficulty. The study continued with a description of preterm infant oral feeding disorder characteristics. The Board Committee on Medical/Health Research Ethics of Universitas Indonesia conducted the ethical review for this research, with protocol number 22–09–1117.

The inclusion criteria included (1) participants with a gestational age of less than 37 weeks at birth, (2) participants with a history of treatment in the studied hospital's NICU, and (3) participants with a corrected age of less than 3 months during the FEES examination. The exclusion criteria for this study included (1) participants with craniomaxillofacial abnormalities, such as cleft lip palate and facial cleft, (2) participants with insufficient medical data, including basic characteristics, clinical characteristics, and morbidity, and (3) participants with videos of FEES examinations that are difficult to assess or incomplete (blurry video image or incomplete FEES stage examination).

Medical records were used to obtain secondary data on birth history, postnatal characteristics, and risk factors for feeding and swallowing disorders. Comorbidities are classified into three categories: central nervous system (CNS) comorbidities (such as intraventricular hemorrhage, periventricular leucoplakia, hydrocephalus, and ventriculomegaly), cardiopulmonary comorbidities (such as bronchopulmonary dysplasia, hyaline membrane disease, pneumonia of prematurity, respiratory distress syndrome, patent ductus arteriosus, and other congenital heart diseases), and gastrointestinal (GI) comorbidities (such as GERD, necrotizing enterocolitis, and Hirschsprung disease). The neonatal medical index (NMI) was used to assess the severity of comorbidities.[13]

Oral−motor examination

Medical records and physical examination videos were used to obtain data on oral motor examination and postural tone. Infant feeding difficulty criteria are based on several components including (1) oral motor assessment using the neonatal oral−motor assessment scale (NOMAS),[14] (2) preterm infant morbidity severity using a neonatal medical index (NMI),[13] and (3) oral feeding readiness and ability score (OFRAS) based on the Luh Karunia Wahyuni (LKW) score.[15]

FEES examination

The FEES examination is used to assess oral feeding difficulties based on the penetration aspiration scale (PAS)[16] and the normal SSB coordination sequence ratio. The FEES examination was performed by assessing bottle-feeding for the SSB sequence ratio examination and a spoonful of milk for assessing the pharyngeal phase. Participants in the normal group had a normal suck: swallow: breath ratio (1 : 1 : 1 to 2 : 2 : 1) without an apnea phase or apnea of less than 2 s, followed by a PAS score of 1−2 in the FEES examination. Participants in the dysphagia group had FEES PAS scores greater than 2 (3 to 8). Participants in the SSB incoordination group were defined as those who could not maintain optimal SSB cycle ratio at 1 : 1 : 1 to 2 : 2 : 1 and/or were followed by a single phase of apnea or pause of breath lasting less than 2 s, despite having a normal PAS score (PAS 1−2).[2,12,17] FEES examination data were obtained from medical records and FEES examination videos. Two bronchoesophagology consultant doctors (EZKR, ST) with more than 15 years of experience in pediatrics evaluated and validated the FEES videos. Blinded validation was performed, and the Kappa value for inter-rater and intra-rater agreement was calculated. A univariate analysis was performed to determine the prevalence, distribution, and characteristics. The Statistical Package for Social Science (SPSS) 20 software was used to analyze the data.

Results

There were 43 preterm infants referred to the Integrated Dysphagia Clinic between October 2020 and October 2022. One baby was excluded due to labiopalatoschizis disorder, and two others were excluded due to incomplete FEES videos. Forty participants fit the research criteria. Validity assessment revealed an inter-rater agreement with a κ value of 0.62−0.636 and an intra-rater agreement with a κ value of 0.625 −0.80 upon residue, penetration, and aspiration assessment. The prevalence of swallowing disorders or dysphagia was 25 %, the prevalence of SSB incoordination was 62.5 %, and the prevalence of infants with normal feeding ability was 12.5 % (Table 1).

Table 1 Distribution of Subject Characteristics Based on Diagnosis of Dysphagia or Suck-swallow-breath (SSB) Incoordination.

Subject characteristics	(*n* = 40)%
Dysphagia	10 (25 %)
Oral Mechanic	1 (10 %)
Pharynx Neurogenic	5 (50 %)
Oropharynx Neurogenic	4 (40 %)
Oral Feeding Disorder	
SSB Incoordination	25 (62.5 %)
Normal oral feeding and swallowing function	5 (12.5 %)

The female gender was dominant at 62.5 % (13/40), the average gestational age was 31.4 ± 3.3 weeks, and the average post-menstrual age (PMA) was 37.6 ± 3.2 weeks. The difference in PMA between dysphagia and SSB coordination disorder was significant. The median duration of mechanical ventilation was 27.5 (0−178) days. The median duration of length of stay (LOS) was 4 (4−22) weeks. The difference in the median duration of LOS was statistically significant (Tables 2 and 3).

The pre-swallowing assessment characteristics found were retropalatal collapse (42.5 %), paradoxical vocal cord movement (PVCM) (15 %), high arched palate (17.5 %), laryngomalacia (47.5 %), and GERD (60 %). The FEES examination characteristics included inadequate NS parameters (65 %), penetration (22.5 %), and aspiration (15 %) (Table 4).

Discussion

The authors found that the prevalence of dysphagia in preterm infants was 25 %, and the prevalence of SSB incoordination was 65 %. According to Motion et al.,[18] the prevalence of dysphagia in preterm infants ranges from 24.5 % to 26 %. Vetter−Laracy et al.[19] and Da Costa et al.[20] found that dysphagia was present in 71 % and 64.2 % of preterm infants, respectively. The difference in prevalence is due to differences in population, study design, and dysphagia evaluation modalities. SSB incoordination often results in desaturation, choking, or pauses in breathing while feeding that clinically mimic symptoms of dysphagia in infants. Because SSB coordination should be assessed via a combination of NOMAS, OFRAS, and FEES examinations, the previous study[18-20] could not distinguish the presence of abnormal SSB cycle integration as a different pathologic phenotype in oral feeding disorder in prematurity owing to the use of different examination methods. The present study is the first to distinguish the pathological conditions of SSB incoordination and dysphagia as the causes of oral feeding difficulties in preterm infants, which have not been previously assessed.

Table 2 Distribution of subjects according to birth characteristics (n = 40).

Birth characteristic	Dysphagia N	SSB incoordination N	Normal N
Sex			
Undefined	1	1	0
Male	1	11	1
Female	8	13	4
Total Male: 32.5 %	Total Female: 62.5 %	Total undefined: 5%	
Gestational age			
23 – 27 weeks	2	3	0
28 – 31 weeks	3	14	3
32 – 36 weeks	5	8	2
Mean+/-SD gestational age	30.9 ± 4.2	31.6 ± 3	31.4 ± 3.2
P-value	0.855[a]		
PMA			
32 – 36 weeks	2	15	2
37 – 41 weeks	3	10	3
42 – 45 weeks	5	0	0
Mean+/-SD PMA	40.2 ± 4	36±2.4	37±1.8
P-value	**0.006**[a]		
Birth Weight			
< 1000	2	12	0
1000 – 1500	2	11	2
1501 – 2500	5	1	2
> 2500	1	1	1
Median birth weight	1825 (600–2600)	1550 (530–4500)	1900 (1170–4200)
Mean rank	20.60	19.42	25.70
P-value	0.547b		
5 min APGAR			
Apgar 0 – 6	5	7	2
Apgar 7 – 10	5	18	3
Median APGAR	6.5 (3–9)	8 (4–10)	9 (4–10)
Mean Rank	20.6	19.42	25.7
P-value	0.994[b]		

PMA, Post-menstrual age; SSB, suck-swallow-breath; APGAR, Appearance, pulse, grimace, activity, respiration.
#One-way ANOVA/Welch test. Post hoc Games-Howell PMA dysphagia vs. SSB Incoordination p = 0.005, dysphagia vs. normal p = 0.111, SSB Incoordination vs. normal p = 0.963.
*Kruskal-Wallis test.

Feeding difficulties in preterm infants are caused by immaturity of anatomical, neuromuscular, and sensorimotor functions and risk factors, such as gestational age, APGAR score, low birth weight, comorbidities, and the use of mechanical ventilation. The present study included the largest number of participants between the ages of 32- and 36-week PMA, with an average gestational age (GA) of 30–31 weeks. Oral motor development, which includes sucking and suck–swallow mechanisms, mostly develops at 30–36 weeks of gestation, along with the development of postural tone.[2,12] While the infant is feeding, postural tone helps support the cervical spine to maintain good posture. Immaturity of this system will jeopardize airway patency during feeding due to the inability of the suprahyoid muscle to strengthen the neck and laryngeal elevation while swallowing.[21] Later, over 37 weeks PMA, infants will develop their self-regulatory and balancing system, which allows coordination of the suck–swallow mechanism with respiratory effort (decreased ventilation rate during feeding), resulting in oral feeding milestones at discharge that are more common in term infants. Thus, intrauterine maturation at birth leads to extrauterine oral feeding maturation. In contrast to infants born at < 30 weeks of gestation, premature infants have not been exposed to oral–motor developmental, postural tone, self-regulation, and balancing systems. Furthermore, several interruptions in breath due to the increased frequency of swallowing apnea, especially in infants < 34 weeks PMA, make it difficult for these infants to coordinate the suck–swallow mechanism with respiratory effort. Apnea caused by swallowing or feeding reduces oxygen saturation, triggering the “diving response” and resulting in bradycardia.[1,2,12,22] The authors found that infants in the present study who had PMA maturity, especially those in the dysphagia group, had an older PMA of ±40 weeks. This was different from those with SSB incoordination, who had an average PMA of ±36 weeks, and there were statistically significant ($p < 0.05$) differences in PMA mean between groups. It has been suggested that multi-system intrauterine immaturity will delay

Table 3 Subject Distribution by postnatal risk (n = 40).

Post-Natal Risk	Dysphagia (N)	SSB Inccordination (N)	Normal (N)
Length mechanical ventilation			
0 − 10 days	4	8	4
> 10 days	6	17	1
Median mechanical ventilation	21.5 (0−178)	29 (1−81)	10 (4−62)
Mean rank	21	21	16.6
P-value	0.727*		
Median Length of stay (weeks)	8 (4−22)	4 (4−10)	4 (4−8)
Mean rank	30.20	16.88	19.20
P-value	**0.004**[a]		
CNS Comorbidity			
Yes	4	10	4
No	6	15	1
Cardiopulmonary Comorbidity			
Yes	8	24	5
No	2	1	0
Gastro-intestinal Comorbidity			
Yes	8	14	1
No	2	11	4
NMI			
4 − 5 (severe)	6	18	2
3 (moderate)	3	5	3
1 − 2 (mild)	1	2	0
Median NMI	4 (1−5)	4 (2−5)	3 (3−5)
Mean rank	20.10	21.20	17.80
P-value	0.809[a]		

CNS, central nervous system; NMI, neonatal medical index; SSB, suck-swallow-breath.

[a] Kruskall−Wallis test. Post hoc Bonferroni test for length of stay (LOS) dysphagia vs. SSB Incoordination p = 0.003, dysphagia vs. normal p = 0.186, SSB Incoordination vs. normal p = 1.0.

extrauterine feeding milestones owing to the absence of intrauterine memory, as reported by Jadcherla et al.,[23] who reported drawbacks for feeding milestones and prolonging the length of stay (LOS) in infants with < 28 weeks PMA, even when they reach extrauterine maturation.

In the present study, the severe morbidity rate was 26 of 40 infants (65 %), with six infants (23 %) having dysphagia, 18 infants (69 %) having SSB incoordination, and only two (8 %) exhibiting normal feeding. The treatment period for the infants ranges from 8 to 26 weeks. Multiple comorbidities in preterm infants play a significant role in their inability to meet feeding milestones. Preterm infants are more likely to have cardiorespiratory, central nervous system, and gastro-intestinal comorbidities.[23] The authors found prolonged use of mechanical ventilation (> 21 days) followed by the highest prevalence of cardiopulmonary comorbidity (92.5 %), especially in the case group. This is closely related to mechanical ventilation dependency and may show a possible correlation between prolonged mechanical ventilation use and a higher morbidity scale, affecting infant oral feeding performance. According to Harding et al.,[24] approximately 50.3 % of patients with delayed feeding development are given a variety of medications while hospitalized. They also found that very preterm infants had a high prevalence of multiple comorbidities; with 31 % facing delayed oral feeding when discharged, and nearly 58 % still requiring speech-language therapy (SLT) support at home. Prolonged use of ventilatory devices in preterm infants due to bronchopulmonary dysplasia (BPD) results in an inability to modulate respiration to follow the suck−swallow rhythm, resulting in desaturation during feeding. Additionally, the longer duration of invasive medication contributes to oral motor restriction and failed maturation of self-regulatory function in postnatal infants, with delayed oral feeding ability.[2,23,24] Moreover, long-term invasive medication around the mouth (i.e., an endotracheal tube, suctioning, or a feeding tube) also contributes to generating oro-sensory aversion owing to noxious sensory stimulation that affects pharyngeal swallowing ability.[25] In the present study, the Bonferroni post hoc test for a median duration of LOS was statistically significant between dysphagia and SSB incoordination. This result supports the possible relationship between the long-term use of invasive medication during hospitalization and the resulting oro-sensory aversion in the form of pharyngeal dysphagia, which was typically observed in the dysphagia group.

Anatomical abnormalities of the upper airway are also a factor that can exacerbate SSB cycle coordination. The present study found the prevalence of laryngomalacia, retropalatal collapse, PVCM, and GERD as disorders can interfere with the coordination of the SSB cycle and can lead to preterm infant swallowing disorders. The authors found that 42.5 % of patients had retropalatal collapse, 15 % had PVCM, 47.5 % had laryngomalacia, 60 % had GERD, and 62.5 % had inadequate NS, which was mostly found in the SSB

Table 4 Oral motor reflex, postural tone examination, and flexible endoscopic evaluation of swallowing (FEES) examination characteristic (*n* = 40).

Examination	Dysphagia N (%)	SSB Incoordination N (%)	Normal N (%)
Rooting Reflex			
Adequate	5 (12.5)	19 (47.5)	5 (12.5)
Inadequate	5 (12.5)	6 (15)	0
Total adequate rooting reflex	29 (72.5)		
Total inadequate rooting Reflex	11 (27.5)		
Sucking Reflex			
Adequate	6 (15)	21 (52.5)	5 (12.5)
Inadequate	4 (10)	4 (10)	0
The total adequate sucking reflex	32 (80)		
The total inadequate sucking reflex	8 (20)		
NNS			
Adequate	2 (5)	15 (37.5)	5 (12.5)
Inadequate	8 (20)	10 (25)	0
Total adequate NNS	22 (55)		
Total inadequate NNS	18 (45)		
Postural Tone			
Adequate	6 (15)	15 (37.5)	5 (12.5)
Inadequate	4 (10)	10 (25)	0
Total adequate postural tone	26 (65)		
Total inadequate postural tone	14 (35)		

FEES Examination	Dysphagia N (%)	SSB Incoordination N (%)	Normal N (%)
Lip sealed			
Adequate	7 (17.5)	12 (30)	5 (12.5)
Inadequate	3 (7.5)	1 (2.5)	0
Retro-palatal Collapse			
Yes	7 (17.5)	9 (22.5)	1 (2.5)
No	3 (7.5)	16 (40)	4 (10)
PVCM			
Yes	0	5 (12.5)	1 (2.5)
No	10 (25)	20 (50)	4 (10)
High Arched Palate			
Yes	4 (10)	2 (5)	1 (2.5)
No	6 (15)	23 (57.5)	4 (10)
Tongue-tie			
Yes	3 (7.5)	4 (10)	0
No	7 (17.5)	22 (55)	5 (12.5)
Standing Secretion			
Yes	9 (22.5)	8 (20)	0
No	1 (2.5)	17 (42.5)	5 (12.5)
Laryngomalacia			
Yes	6 (15)	11 (27.5)	2 (5)
No	4 (10)	14 (35)	3 (7.5)
GERD			
Yes	10 (25)	14 (35)	0
No	0	11 (27.5)	5 (12.5)
Laryngomalacia			
Type 1	5	10	2
Type 1 and 3	1	1	0
NS			
Adequate	0	9 (22.5)	5 (12.5)
Inadequate	10 (25)	16 (40)	0

Table 4 (*Continued*)

FEES Examination	Dysphagia N (%)	SSB Incoordination N (%)	Normal N (%)
Residue			
Yes	1 (10)		
No	9 (90)		
Penetration			
Yes	9 (90)		
No	1 (10)		
Aspiration			
Yes	6 (60)		
No	4 (40)		

SSB, suck-swallow-breath; NNS, non-nutritive sucking; PVCM, paradoxical vocal cord movement; GERD, gastroesophageal reflux disease; NS, nutritive sucking.

incoordination group. Only GERD was observed as an absolute comorbidity in the dysphagia group. Laryngomalacia is frequently associated with the presence of neuromuscular disease in preterm infants, along with a higher prevalence of GERD, which will alter the sensorimotor integration of the LAR. Acid reflux causes LAR dysfunction, which results in an incorrect signaling pattern via the superior laryngeal nerve (SLN) and alters laryngeal function, such as decreased laryngeal tone, choking, aspiration, and inability to clear secretion. This will jeopardize upper airway patency, resulting in uncoordinated respiratory, aspiration, and desaturation. In special cases, very preterm infants with immature sensorimotor integration of the LAR will experience LAR hyperactivity, which will increase the acceleration of glottic closure opposite to inspiration.[4,26] When moving, the turbulence of air passing through the vocal cords paradoxically produces a high-pitched stridor sound when inspired (Munchausen stridor), known as PVCM.[27] This condition also contributes to abnormal respiratory effort in preterm infants.

The characteristics observed in the oral motor examination and FEES also contribute to differentiating dysphagia in preterm infants who have SSB incoordination. In the dysphagia group, inadequate NNS and NS ability dominate with a higher PAS score, which represents the lack of sucking and swallowing ability due to oro-sensory aversion. However, the SSB incoordination group showed a prevalence of inadequate NS, and some had adequate NNS ability. The different physiological characteristics between NNS and NS underlay this condition in which NS requires coordination between suck−swallow and respiratory effort to maintain a rhythmical SSB cycle during oral feeding.[25,28]

The clinical goal of this study was to differentiate the distinct phenotypes between dysphagia in preterm infants and SSB incoordination. The previously mentioned condition describes when preterm infants experience prolonged feeding difficulties after being discharged home. Bertoncelli et al.[29] and Lau et al.[25] considered early intervention to resolve feeding problems in preterm infants. Sensorimotor intervention, such as NNS stimulation with a pacifier and swallowing exercises (SEs), will stimulate the integration of the optimal alert state with sensorimotor function in the oral, tactile, and kinesthetic domains. Robust alertness before and during feeding, however, will improve infant feeding competence and improve the quality of sucking and the number of suckings per burst. This intervention will improve oral feeding performance as the infant matures. Another method proposed by Ross and Philbin[22] is a cue-based bottle-feeding intervention known as Supporting Oral Feeding in Fragile Infants (SOFFI). The infant must be physiologically ready for oral feeding before the caregiver observes the infant's "cue" during feeding, such as physiologic instability, lack of engagement in feeding, and SSB cycle integration difficulty. Cue-based feeding intervention will allow us to provide an advanced feeding strategy when the infant is ready to be discharged; this could be full-demand feeding, semi-demand feeding, or full-gavage feeding. Despite the results, the present study did not exclude infants who had previously performed oral motor exercises, and long-term hospitalization became a potential bias that could affect the assessment of oral feeding and swallowing in this study.

Conclusion

Several risk factors could result in differences between dysphagia and SSB incoordination in preterm infants. Dysphagia in preterm infants is mostly observed in those with mature PMA, longer LOS, and the presence of GERD with inadequate NNS and NS abilities. SSB incoordination is primarily observed in those with immature PMA, a higher prevalence of cardiopulmonary comorbidity, and a higher prevalence of upper airway pathologies (laryngomalacia, PVCM) with inadequate NS ability.

Conflicts of interest

The authors declare no conflicts of interest.

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Supplementary materials

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References

1. Lau C, Smith EO, Schanler RJ. Coordination of suck-swallow and swallow respiration in preterm infants. Acta Paediatr. 2003;92:721–7.
2. Gewolb IH, Vice FL. Maturational changes in the rhythms, patterning, and coordination of respiration and swallow during feeding in preterm and term infants. Dev Med Child Neurol. 2006;48:589–94.
3. Rommel N, De Meyer AM, Feenstra L, Veereman-Wauters G. The complexity of feeding problems in 700 infants and young children presenting to a tertiary care institution. J Pediatr Gastroenterol Nutr. 2003;37:75–84.
4. Thompson DM. Abnormal sensorimotor integrative function of the larynx in congenital laryngomalacia: a new theory of etiology. Laryngoscope. 2007;117:1–33.
5. Aliyu I, Lawal T, Onankpa B. Hypoxic-ischemic encephalopathy and the Apgar scoring system: the experience in a resource-limited setting. J Clin Sci (Philadelphia). 2018;15:18–21.
6. Jadcherla S. Dysphagia in the high-risk infant: potential factors and mechanisms. Am J Clin Nutr. 2016;103:622S–8S.
7. Carvalho CG, Silveira RC, Procianoy RS. Ventilator-induced lung injury in preterm infants. Rev Bras Ter Intensiva. 2013;25:319–26.
8. Armstrong ES, Reynolds J, Sturdivant C, Carroll S, Suterwala MS. Assessing swallowing of the breastfeeding NICU infant using fiberoptic endoscopic evaluation of swallowing: a feasibility study. Adv Neonatal Care. 2020;20:244–50.
9. Miller CK, Willging JP. Fiberoptic endoscopic evaluation of swallowing in infants and children: protocol, safety, and clinical efficacy: 25 years of experience. Ann Otol Rhinol Laryngol. 2020;129:469–81.
10. Reynolds J, Carroll S, Sturdivant C. Fiberoptic endoscopic evaluation of swallowing: a multidisciplinary alternative for assessment of infants with dysphagia in the neonatal intensive care unit. Adv Neonatal Care. 2016;16:37–43.
11. Als H. Toward a synactive theory of development: promise for the assessment and support of infant individuality. Infant Ment Health J (New J). 1982;3:229–43.
12. McCain GC. An evidence-based guideline for introducing oral feeding to healthy preterm infants. Neonatal Netw. 2003;22:45–50.
13. Korner AF, Stevenson DK, Kraemer HC, Spiker D, Scott DT, Constantinou J, Dimiceli S. Prediction of the development of low birth weight preterm infants by a new neonatal medical index. J Dev Behav Pediatr. 1993;14:106–11.
14. Osman A. Oral feeding readiness and premature infant outcomes. J Neonatal Nurs (New J). 2019;25:111–5.
15. Wahyuni LK, Mangunatmadja I, Kaban RK, Rachmawati EZ, Harini M, Laksmitasari B, et al. Factors affecting oral feeding ability in indonesian preterm infants. Pediatr Rep. 2022;14:233–43.
16. Butler SG, Markley L, Sanders B, Stuart A. Reliability of the penetration aspiration scale with flexible endoscopic evaluation of swallowing. Ann Otol Rhinol Laryngol. 2015;124:480–3.
17. Lau C. Development of suck and swallow mechanisms in infants. Ann Nutr Metab. 2015;66:7–14.
18. Motion S, Northstone K, Emond A. The ALSPAC Study Team. Persistent early feeding difficulties and subsequent growth and developmental outcomes. Ambul Child Health (New J). 2001;7:231–7.
19. Vetter-Laracy S, Osona B, Roca A, Peña-Zarza JA, Gil JA, Figuerola J. Neonatal swallowing assessment using fiberoptic endoscopic evaluation of swallowing (FEES). Pediatr Pulmonol. 2018;53:437–42.
20. Da Costa MA, Krüger E, Kritzinger A, Graham MA. Prevalence and associated prenatal and perinatal risk factors for oropharyngeal dysphagia in high-risk neonates in a South African hospital. S Afr J Commun Disord. 2019;66:e1–8.
21. Drake E. Positioning the Neonate for Best Outcomes. Glenview, IL: National Association of Neonatal Nurses; 2017.
22. Ross ES, Philbin MK. Supporting oral feeding in fragile infants: an evidence-based method for quality bottle-feedings of preterm, ill, and fragile infants. J Perinat Neonatal Nurs. 2011;25:349–57. quiz 358-9.
23. Jadcherla SR, Wang M, Vijayapal AS, Leuthner SR. Impact of prematurity and co-morbidities on feeding milestones in neonates: a retrospective study. J Perinatol. 2010;30:201–8.
24. Harding C, Bell N, Griffiths S, Michou E. A descriptive evaluation of early feeding development of infants in a local neonatal unit. J Neonatal Nurs. 2023;29:681–6.
25. Lau C, Smith EO. Interventions to improve the oral feeding performance of preterm infants. Acta Paediatr. 2012;101: e269–74.
26. Ayari S, Aubertin G, Girschig H, Van Den Abbeele T, Mondain M. Pathophysiology and diagnostic approach to laryngomalacia in infants. Eur Ann Otorhinolaryngol Head Neck Dis. 2012;129:257–63.
27. Andrianopoulos MV, Gallivan GJ, Gallivan KH. PVCM, PVCD, EPL, and irritable larynx syndrome: what are we talking about and how do we treat it? J Voice. 2000;14:607–18.
28. Ostadi M, Jokar F, Armanian AM, Namnabati M, Kazemi Y, Poorjavad M. The effects of swallowing exercise and non-nutritive sucking exercise on oral feeding readiness in preterm infants: a randomized controlled trial. Int J Pediatr Otorhinolaryngol. 2021;142:110602.
29. Bertoncelli N, Cuomo G, Cattani S, Mazzi C, Pugliese M, Coccolini E, et al. Oral feeding competences of healthy preterm infants: a review. Int J Pediatr. 2012;2012:896257.