

# **DEVELOPMENT AND VALIDATION OF A SCREENING TOOL FOR DETECTION OF DYSPHAGIA IN NEONATES**



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## **Principal Investigator**

Dr. N. Swapna

Associate Professor of Speech Pathology  
Department of Speech-Language Pathology

## **Research officer**

Ms. Seshasri D

**ALL INDIA INSTITUTE OF SPEECH AND HEARING  
MANASAGANGOTTHRI, MYSURU - 570 006**



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**Research officer**  
Ms. Seshasri. D

**Principal investigator**  
Dr.Swapna. N

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# CHAPTER I

## Introduction

Oral feeding is the most complex sensori-motor process in the new-borns. It requires suckling and swallowing along with co-ordination in the breathing. The primary measure of successful feeding is adequate growth, in terms of weight gain in early infancy as well as during the first few years of life (Arvedson, 2006). Successful and efficient oral feeding requires many factors such as adequate oral sensorimotor control and swallowing skills, overall adequate health (including respiratory and gastrointestinal function), neural maturation, and musculoskeletal tone.

The feeding in infants can be broken down into different events. The oral phase, which includes suckling and the transportation of the fluid towards the pharynx, triggering of the swallowing reflex, pharyngeal phase, which involves the transportation of the fluid through the pharynx and esophageal phase, in which the fluid is transported through the esophagus to the stomach. However, in certain infants due to various causes, the feeding and swallowing ability can get compromised leading to dysphagia. Dysphagia occurs when there is any disruption in any of the events mentioned above, which compromises the safety efficiency of swallow or adequacy of nutritional intake. The synchrony between breathing and swallowing is also equally important for successful swallowing. Because swallowing and breathing share a common space in the pharynx, problems in either of these processes, or lack of synchronization between these processes, can affect the ability to protect the airway during swallowing (Doddrill, 2014).

Dysphagia can occur either in the oral, pharyngeal or esophageal phases. At times, problems can be seen in the oropharyngeal phase and/or the pharyngo-esophageal phase. The

presenting symptoms and signs observed in the oropharyngeal phase include latching problems, delay in suck, lack of rhythm and lingual movement, poor extraction of bolus, nasopharyngeal regurgitation, delayed initiation of pharyngeal swallow, silent aspiration, peristaltic failure and gagging. The symptoms and signs observed in the pharyngo-esophageal phase include pharyngeal pooling, wet gurgly breathing, cough with feedings, stridor, nasopharyngeal regurgitation, delayed pharyngeal phase, pharyngo-UES dyscoordination, laryngeal penetration, laryngeal aspiration, apnea, bradycardia and desaturations, and cardiorespiratory events. Other potential symptoms of dysphagia include back arching while feeding, reduced responsiveness, weak sucking, fatigue, shorter (less than 5 minutes) or prolonged period (more than 30 minutes) of feeding, respiratory problems during or after feeding, food aversion, anterior spillage of fluid, wet/gurgly voice immediately after feeding, weight loss and failure to thrive (Jadcherla, 2016; Farneti & Genovese, 2017). If these difficulties in neonates are left untreated, it can lead to increased rates of morbidity and mortality, particularly among neonates with life-threatening medical conditions (Bae, Lee, Seo, Oh & Han, 2014).

Dysphagia can occur frequently in neonates due to associated medical conditions such as prematurity, cardiopulmonary diseases and neurological disorders (Jadcherla, 2016). Neonatal dysphagia is commonly reported in new-borns with very low birth weight (less than 1500 grams) and extremely preterm (28 to 32 weeks) neonates. Schoeman and Kritzinger (2017) reported that low birth weight and prematurity are the risk factors commonly associated with neonatal dysphagia, and further claimed that these neonates with feeding difficulties had multiple medical conditions than a single risk factor. Other risk factors associated with neonatal dysphagia include congenital anomalies, peri-natal asphyxia, gastrointestinal disorders, post-surgical and sepsis



categories. Such risk factors disrupt feeding process and the consequences of these affected feeding processes carry over into infancy and toddler age groups (Jadcherla, 2016).

The incidence of feeding and swallowing disorders is increasing because of improved survival rates of children with complex and medically fragile conditions (Lefton-Greif, 2008; Lefton-Greif, Carroll, & Loughlin, 2006; Newman, Keckley, Petersen, & Hamner, 2001) and the improved longevity of persons with dysphagia that develops during childhood (Lefton-Greif, McGrattan, Carson, Pinto, Wright, & Martin-Harris, 2017). Estimated reports of the incidence and prevalence of paediatric feeding and swallowing disorders vary widely due to factors including variations in the conditions and populations sampled, the way feeding disorders and/or swallowing impairment are defined, and the choice of assessment methods and measures (Arvedson, 2008; Lefton-Greif, 2008). An estimated 116,000 new-born infants are discharged from short-stay hospitals with a diagnosis of feeding problems, according to the National Hospital Discharge Survey from the CDC (National Centre for Health Statistics, 2010). The prevalence of feeding problems in premature infants born at <37 week of gestation is ~10.5%, and this frequency increases to ~24.5% among those born with a very low birth weight (<1500 g) (Motion, Northstone, & Emond, 2001). Twenty to eighty percent of high-risk neonatal intensive care unit infants have feeding concerns during infancy (Field, Garland, & Williams, 2003; Rommel, De Meyer, Feenstra, & Veereman-Wauters, 2003).

Therefore, to avoid various complications, dysphagia must be diagnosed as early as possible. Early detection of feeding and swallowing difficulties in neonates plays an essential role in preventing and reducing the associated developmental and medical complications and their negative impact on both the child and caregivers (Prasse & Kikano, 2009). The early identification of dysphagia is vital to determine a suitable treatment plan to support adequate

nutrition and growth (Lefton-Grief, 2008). However, due to the involvement of various physiological systems, feeding and swallowing difficulties in these vulnerable populations are very complex in nature (Arvedson, 2008; Bruns & Thompson, 2012; Jadcherla, 2016) and challenging to assess.

Screening tools are widely used to decide on the presence or absence of dysphagia because they can be easily administered without rigorous training, are non-invasive, quickly interpretable, time and cost-effective. Screening using a validated tool is an important part of early identification of feeding and swallowing difficulties in neonates (Thoyre, Park, Pados, & Hubbard, 2013). The main objective of any dysphagia screening tool is to identify its presence as early as possible, so as to make referral for detailed evaluation, followed by the formulation of an appropriate intervention plan (Leder&Espenosa, 2002; Duarte, 2010). Further, questionnaires are widely adopted method among the various screening methods to classify and characterize the signs and symptoms of any condition (Orenstein, 2006).

### **Need for the study**

A systematic literature review revealed that although there are many assessment scales and checklists, there are limited screening tools for detecting dysphagia, especially in neonates. Although the recognition of neonatal dysphagia and its challenges has increased (Jadcherla, 2016), research into the development of clinically validated screening tools for this population continues to remain limited (Heckathorn, Speyer, Taylor, & Cordier, 2016).

The available screening tools include Dysphagia Screening Test for pre-term infants (DST-PI, Lee & Seo, 2017), Feeding and Swallowing Scale for Premature Infants (FSSPI, Moon, Jung, Cheon, Oh, Ki, & Kwon, 2017) and Infant and Child Feeding Questionnaire” (ICFQ)

(Byrd, Steinfeld, Hoffmann, & Silverman, 2017). Most of the tools have been developed for neonates with specific issues, for example, DST-PI and FSSPI have been designed for pre-term infants. Tools that can be used across all neonates, irrespective of the underlying high risk factor are limited. Though DST-PI and FSSPI are validated tools, the ICFQ is yet to be validated.

Feeding difficulties are considered as potential markers for neonatal brain injury (Jadcherla, 2016). Hence, identification of feeding difficulties paves way for the identification of brain damage. This facilitates early detailed assessment followed by an accurate diagnosis and initiation of intervention, so as to reduce or prevent its negative sequelae, which include inadequate weight gain, dehydration, and limited oral sensory experience, which may continue to impact on infancy and early childhood. Early identification and early intervention of feeding and swallowing problem could contribute to a better quality of life in the neonate and the family members. In addition, the information obtained on early detection of dysphagia through screening can also help the professionals to educate the parents/caregivers on the need to address the difficulties associated with feeding and swallowing as early as possible.

Further in 2003, the World Health Organization (WHO) and UNICEF adopted the “Global strategy for infant and young child feeding” and identified that different feeding practices exist worldwide from birth to infancy which is related to their culture, socio-economic status and educational backgrounds, therefore region specific practices should be given importance during evaluation and management of feeding disorders in infants. Thus, there is a need to develop a context specific, validated screening tool to assess dysphagia in neonates, which would further facilitate management. This would facilitate the speech pathologists to evaluate these infants and make informed decisions on the appropriate management of dysphagia in high risk neonates.

In many developing countries such as India, considering the limited resources and shortage of equipment to conduct objective evaluation, particularly in rural areas, to detect and diagnose dysphagia in neonates, it is essential that easy and quick to administer screening tools are developed. Considering the high prevalence of neonatal dysphagia and its adverse impact on quality of life and the lack of screening tools with well-established psychometric properties, scoring and interpretation algorithm for early detection of neonatal dysphagia, a need was felt to develop a screening tool for the early identification of dysphagia in the neonatal population.

### **Aim of the study**

The principle aim of this study was to develop and standardize a screening tool for early identification of dysphagia or any feeding and swallowing difficulties in the neonatal population with high risk factors.

### **Objectives of the study**

The specific objectives of the study were

1. To construct the feeding and swallowing screening tool for early identification of dysphagia in neonates by selecting and including appropriate test items.
2. To establish the content validity of the developed tool based on the multidisciplinary expert input.
3. To clinically validate the developed tool by administering it on the typical, high risk and very high-risk neonates.
4. To establish the sensitivity and specificity of the developed tool by a retrospective assessment of feeding and swallowing difficulties on another known population of children with and without dysphagia.

5. To verify the inter-rater reliability of the developed tool.
6. To identify the critical test items of the developed tool in order to develop a shorter version of the tool.
7. To establish the scoring and interpretation algorithm of the developed tool.

## CHAPTER II

### Review of Literature

Feeding and swallowing are essential for survival, growth and development, nutrition and overall well-being of neonates and young infants. Feeding has primary importance within the living experience of the neonates and it continues to be a major binding element of experience in the lives of infants and children. Also, it is a sensori-motor skill that matures during the first two years of life to provide nutrition for normal growth and development. The satiation of hunger and homeostasis is achieved through the feeding process. It also provides opportunity for oral sensory and motor stimulation, mother-child bonding and oromotor skill development (Kummer, 2008). According to Arvedson and Brodsky (2002), feeding is the process involving any aspect of eating/drinking including gathering and preparing food for intake, sucking (for liquids) or chewing (for solids) and swallowing. That is, feeding is the process of eating or the act of being fed and involves the process of swallowing.

Swallowing (also referred as deglutition in some scientific context) is the process that allows the food substances to pass from mouth to the esophagus via pharynx (Farneti& Genovese, 2016). Swallowing is a very complex process and involves four phases such as oral preparatory, oral transit, pharyngeal and oesophageal phase. In the oral preparatory phase, food is manipulated with saliva to form a cohesive bolus. It is completely a voluntary phase which includes sucking liquids, chewing solid food and even manipulating soft boluses. Oral transit phase involves voluntarily propelling the cohesive bolus posteriorly by the tongue. It results in the initiation of pharyngeal phase of swallowing. During pharyngeal phase which is involuntary, the bolus is further propelled by the peristaltic contractions of the pharyngeal constrictors

through the pharynx. In the esophageal phase, the bolus is further carried through the esophageal peristaltic process to the stomach, involuntarily, for digestion.

Eating and swallowing functions are directed by complex neurological system through more than thirty nerves and muscles (Matsuo& Palmer, 2008). Neural swallowing control involves several regions of the central nervous system extending from the motor nuclei of the brainstem to the cortex. Voluntary initiation of swallowing requires bilateral activation of frontal, prefrontal and parietal area. These frontal swallowing areas of the cortex project via descending pathways to medullary swallowing centres in the brainstem. In addition to the cortical regions, brainstem also plays a vital role in achieving the complex coordinated swallowing activity (Mistry &Hamdy, 2008). In the brainstem structures, the clusters of swallowing-related neurons are formed as a central swallowing pattern generator which is deeply connected with the nucleus tractus solitarius in organising the swallowing functions (Bieger&Neuhuber, 2006). These complex co-ordinated neurological activities form the vital base which in turn permits the safe oral transport of food substances for digestion.

The development of feeding and swallowing is an extremely complex and rapid process during the first year of life. This complexity is because of the influence of multiple factors such as structural integrity, growth, neurophysiologic maturation, learning, social maturation and various other environmental and cultural factors. An understanding of these complex developmental milestones of feeding and swallowing abilities is very essential. It enables the health care professionals to detect and provide appropriate guidance on managing the feeding and swallowing difficulties as well as to direct the parents/caregivers to the appropriate professionals (Stevenson & Allaire, 1991).

## **Development of oral feeding skills**

To distinguish between normal physiology and pathology for early detection, understanding of the development of oral feeding skills is important. Feeding and swallowing development requires a highly complex array of interactions starting in embryological and foetal phases and continuing into infancy and early childhood (Delaney & Arvedson, 2008).

The early development of swallowing and oro-motor control has been documented in foetal in-utero studies. Swallowing in utero is essential for controlling the amount and composition of amniotic fluid, re-circulating solutes in the uterus and for the maturation of gastro-intestinal tract of the foetus. In pharynx, one of the first motor responses is pharyngeal swallow which has been observed between 10 to 12 weeks of gestation (Devries, Visser & Prechtel, 1985). However, recent studies have shown this pharyngeal swallowing by 15 weeks of gestation in most foetuses and consistently observed by 22 to 24 weeks of gestation (Miller, Sonies, & Macedonia, 2003).

The distinct backward and forward tongue movements (suckling) starts around 18<sup>th</sup> to 24<sup>th</sup> week of gestation and its frequency can be modified by taste as taste buds become evident at 7<sup>th</sup> week of gestation. Further in suckling, backward movement of the tongue tends to be more pronounced than the forward movement because it does not move beyond the lip borders. At 28 weeks of gestation, tongue cupping is seen. The frequency of the suckling movements increases in the later gestational period. By 34 weeks of gestation a stable preterm baby is likely to suck and swallow which is well enough to maintain adequate nutrition to oral mode (Arvedson, 2006). Any decrease in the rates of suckling by the foetus may be associated with neurological damage or obstruction in the digestive tract.



Just after birth and during the first few months of life, the child's nutritional needs are primarily met by the mother's breast milk or formula milk. Feeding in this stage is reflexive in nature and the neonates are only able to suckle (suckling reflex consists of forward and backward movement patterns of the tongue) and swallow. This reflexive suckling is mediated under brainstem control (Arvedson& Brodsky, 1993). In addition, the most complex sensori-motor process the new-born child undertakes is oral feeding which requires co-ordination among suckling, swallowing, and breathing which continues to refine over the first year of life. This suck-swallow-breathe pattern depends on various factors such as brainstem control, adequate myelination, sensory input and motor performance (Barlow, 2009). The development of oral feeding skills depends not only on the suck-swallow-breathing sequence, but also on various other factors during feeding such as feeder's positional support to the child, mother-child interaction, the ability of mothers to recognize hunger and satiety cues for responsive feeding, as well as the reduction of stress signals of the child (Arvedson& Brodsky, 2002; Lau, 2015).

Further, from birth till 4 months, young infants consume only fluids (breast / formula milk) with complete postural support from the feeder. These young infants primarily rely on primitive oral reflexes for certain actions involved in feeding process. For example, rooting reflex helps them to locate the source of food, suckling reflex to ingest the food being feed and so on. Also, they have certain protective primitive oral reflexes especially for solid foods to prevent it from entering into the airway such as strong gag reflexes, phasic bite and tongue protrusion reflex (Arvedson& Brodsky, 2002).

At 4 to 6 months of age, infants tend to show a gradual transition from forward and backward movements of the tongue (suckle) to up and down movements of the tongue (suck). Also, they explore the oral cavity by bringing their hand to mouth which in turn indicates the

separated movements of the jaw and tongue. These oral exploratory activities by infants help them to desensitize the primitive protective oral reflexes. At this stage, children begin to consume puree consistency and also progress from being entirely dependent on the feeder to semi-dependency where they can make some choices related to the process of feeding (Dodrill, 2014).

Gradually in young infancy, solid diets are introduced to supplement milk based feeds. This transition from liquids to solid food consistency acts as an important biologic marker in the development of feeding. It also enables the child to consume larger variety and volume of liquid and solid based nutrients in order to meet their dietary requirements, which expands as they grow. The child's growth is the salient indicator of their feeding abilities and nutritional intake (Arvedson&Brosdsky, 2002a; Dodrill, 2014). These modifications in the feeding process are the result of neurological maturation in which the higher cortical centres gains more control on feeding and swallowing process than the brainstem.

At 7 to 9 months of age, the infant's diet consists majorly of fluids along with mashed and soft solid foods. They begin to drink from cup along with breast or bottle. As infant grows, some of the primitive oral protective reflexes diminishes (strong gag reflex) and other reflexes like phasic bite reflex are gradually integrated into a voluntary complex oral skill like chewing. In this stage infants begin to sit with semi-dependence (less external support) and also begin assisted self-feeding as the hand to mouth co-ordination improves (Birch & Fisher, 1995; Morris & Klein, 2000).

At 9 to 12 months of age, infants sit independently and begin to stand and walk along with certain fine motor skill. As the nutritional requirements of the infant increases, they begin to

consume mixed proportion of solid and liquid diets (ground and course puree) with greater volume of fluid intake from the cup compared to breast or bottle. Infants in this age develop increased stability and lateralization skills of the jaw and tongue respectively, which in turn improves the efficiency of biting and chewing skills on soft solid food consistency. Also at this stage, the children become fully independent by learning to feed by themselves (Carruth & Skinner, 2002).

From 1 to 2 years of age, toddlers begin to consume greater variety of food of all consistencies, which indicates their refined and coordinated oral feeding abilities (including vertical and lateral mastication movements of the tongue). This in turn increases the mealtime efficiency by enabling them to consume even hard solid food consistencies with little assistance in eating and using utensils (Morris & Klein, 2000; Arvedson& Brodsky, 2002; Dodrill, 2014).

### **Responsive feeding using feeding cues**

Mother and child feeding relationships are a very complex interaction. It requires mothers' necessary responsiveness by interpreting hunger and satiety cues from their newborns (Satter, 1986). This feature is a key dimension of responsive feeding and plays a very significant role in promoting adequate growth and development of the child (Engle, Bentey&Pelto, 2000).

The most common hunger cues were mouthing, rooting and enhanced oral movements/sucking, whereas signs of satiety appeared to be becoming sleepy, decreasing swallowing paces, refusing to eat/detach the nipple, turning the head away and taking more interest in the environment than food (Korner, Chuck, & Dontchos,1968; Crow, 1977; Morris, Rogers, & Taper, 1983; Hodges, Hughes, Hopkinson & Fisher, 2008).

According to Lamb and Easterbrooks in 1981, responsive feeding requires three important aspects in reaction to the child's hunger and satiety cues such as perception, accurate interpretation and appropriate response. In the context of feeding, "perception" refers to the mother's recognition of the child's signals. "Accurate interpretation" indicates an appropriate comprehension of cues exhibited by the child i.e. hunger and satiety cues are interpreted to be a hunger and satiety cues and not as a cue of discomfort or desire to play. Subsequently "appropriate response" requires feeding initiation and termination by the mothers in response to the hunger and satiety cues displayed by their child respectively (Gross, Fierman, Mendelsohn, Chiasson, Rosenberg, Scheinmann, & Messito, 2010). Communication between the mother and the neonate is necessary to promote more effective responsive feeding that leads to a healthy bond that fosters trust (Hotelling, 2004).

However, most mothers are not aware of these natural early feeding cues, particularly hunger cues and some of them believe that hunger is signalled by crying only. According to Worobey, Lopez, and Hoffman in 2009, children should be fed when showing hunger signs and the feeding session should be ended if satiety cues are displayed as opposed to following a scheduled routine. When the child's early feeding cues go unnoticed by the mother, it leads to a breakdown of trust between them and can have long-term effects on the child's well-being and feeding. This leads to maternal dissatisfaction. Satisfaction of the mother with respect to feeding their children is another key aspect of mother and child relationship. The perception of the neonate's progress and satisfaction with feeding by the mother motivates her to continue it effectively. The competence of the neonate with feeding appears to serve as the predominant reinforcer (Matthews, 1991).

## **Different methods of feeding**

Many new-born babies are able to breastfeed immediately after birth without any difficulties. However, in the first few days of life, some babies may not be able to accept breast feeding. In these cases, certain alternative ways to feed the baby must be sought. If breastfeeding is not possible, other methods of feeding can be used to feed a baby such as bottle feeding, paladai/bondla feeding, spoon feeding, cup feeding and tube feeding. Bottle and spoon feeding are the practice of feeding a baby with milk from a feeding bottle and the spoon respectively. Tube feeding is another way of satisfying the nutritional needs of the child, if the oral mode of feeding is not a possibility. Only liquid food consistencies can be used in tube feeding through flexible tubes of different types such as Naso-gastric (NG), Naso-jejunal (NJ), feeding tubes for gastrotomy and jejunotomy.

Of all, paladai feeding is the one that is commonly used in Indian culture especially in south India (Malhotra, Vishwambaran, Sundaram,& Narayanan 1999). A paladai is a small cup with a long, pointed tip that is usually used in some cultures to feed babies with low birth weight. The benefits of using this method of feeding method are less spillage and faster rate of feeding than feeding in spoon or cup. The main drawback of this feeding method is that the feeder should be careful not to spill large quantities of milk into the child's mouth (Marofi, Abedini, Mohammadizadeh&Talakoub, 2016). Figures 2.1 and 2.2 show the images of paladai used in Indian culture and demonstration of feeding a neonate with feeding respectively.



Figure 2.1: Paladai (Source: <https://shopee.com.my/Cup-Feeding-Paladai-i.35351631.523659443>)



Figure 2.2: Feeding a neonate with paladai (Source: <https://shopee.com.my/Cup-Feeding-Paladai-i.35351631.523659443>)

The main objective of feeding is to acquire sufficient nutrients for optimum growth and development. Safe and effective feeding through oral modality in the neonates relies on appropriate development of sucking and swallowing along with breathing co-ordination. Further, normal feeding and swallowing are also dependent on structural integrity of the aero digestive system, neurological maturation along with social interaction and learning, which are influenced by sensory aspects such as oral sensation, motor aspects such as fine and gross motor development and other experimental opportunities. Any disruption to these normal aspects of feeding and swallowing development leads to dysphagia or feeding and swallowing difficulties.

## **Dysphagia**

Problems in any one or more of these four swallowing phases such as oral preparatory, oral transport, pharyngeal and oesophageal phase of swallowing can lead to difficulty in swallowing or dysphagia (Arvedson, 2006). Dysphagia is defined as any disruption to the swallow sequence that result in a compromise of the safety, efficiency and adequacy of nutritional intake (Dodrill&Gosa, 2015). It refers to any problem affecting various stages of swallow leading to nutritional compromise (Jadcherla, 2016).

Children with specific deviation/delays in the developmental processes due to certain medical conditions are at risk for developing dysphagia (Prasse&Kikano, 2009). High-risk status depends on the existence of anything that disrupts the ability of the neonate to engage typically in an expected activity such as feeding. The term "high-risk neonate" refers to neonates having difficulty in oral feeding and swallowing due to medical conditions such as prematurity, respiratory distress syndrome (RDS) and so on (Jadcherla, 2016; Rossetti, 2001). The label "neonatal dysphagia" is commonly used to refer to the feeding and swallowing difficulties that are experienced by high-risk neonates in the period they may stay in neonatal intensive care unit (NICU). This duration can extend beyond the typical neonatal period which is approximately 28 days.

In the literature, two types of dysphagia have been distinguished; oro-pharyngeal dysphagia (a combination of difficulties in the oral and pharyngeal stages of swallow) and esophageal dysphagia (linked to esophageal difficulties) (Jadcherla, Stoner, Gupta, Bates, Fernandez, Di Lorenzo &Linscheid, 2009). Oropharyngeal dysphagia together with esophageal dysphagia represents the major types of dysphagia in neonates.

Neonatal oro-pharyngeal dysphagia refers to any interference with the acts of feeding and/or swallowing that interrupts the oral or pharyngeal stage of swallowing compromising the development of typical feeding and swallowing skills and the neonate's nutritional and respiratory status (Arvedson, 2008; Browne & Ross, 2011; Rogers & Arvedson, 2005). The condition is typically only diagnosed from 32 weeks gestational age when nutritive sucking (NS) should emerge (Rogers & Arvedson, 2005; Thoyre, 2007).

### **Signs and symptoms of neonatal dysphagia**

Neonatal dysphagia is often related to weak sucking, weakness of oral muscles, in-coordination of sucking, swallowing and breathing which can lead to choking, apnea, increased respiratory rate, loud breathing noises, bradycardia / tachycardia, cyanosis, failure to thrive, oxygen de-saturation, aspiration, and a low sugar level (hypoglycaemia) (Laitman & Reidenberg, 1993).

Aspiration is the most important sign of feeding and swallowing difficulties which occurs due to in-coordination among breathing, sucking and swallowing sequence. It may present with/without coughing and choking and over the time and leads to bronchiectasis. Aspiration is the indication of compromised protective reflexes of the larynx/airway. If sensations in the laryngo-tracheal region are also affected, then aspiration can occur without any external sign which is called silent aspiration. The incidence of aspiration ranges from 25% to 73% for infants with swallowing dysfunction (Mercado-Deane, Burton, Harlow, Glover, Dean, Guill, & Hudson, 2001); ~85% of children exhibiting deep laryngeal penetration eventually aspirated, and aspiration occurred 15 s after laryngeal penetration (Newman et al., 2001).



Feeding intolerance is usually reported in new-borns which refer to the difficulty or inability of the subject in digestion or ingestion of the milk fed (Morris & Klein, 2000). It is a commonly encountered in preterm neonates, due to immaturity of the gastro-intestinal functions.

Other commonly observed clinical manifestations of feeding and swallowing difficulties in neonates are back arching, which induces difficulty in positioning the child for feeding, reduced responsiveness, difficulty in initiating sucking and swallowing, shorter (less than 5 minutes) or longer (more than 30 minutes) duration of feeding, refusal shown to the food source by turning the head away from it or by some facial grimaces, labial spillage or anterior spillage of liquid, nasal regurgitation, changes in the voice quality during or after feeding, gagging and frequent vomiting (Jadcherla, 2016).

Moreover, weak sucking in the neonatal period often leads to supplementary tube feeding a using NG or gastroonomy tube, which hampers the development of oral feeding. Hyponatremia, dehydration, hypoglycaemia, hyperbilirubinemia/jaundice and excessive weight loss are the major medical conditions which occur due to inadequate breast feeding. Dehydration leads to serious complications such as cerebral oedema, seizures, intracranial haemorrhage, disseminated intravascular coagulation (excessive clotting or bleeding throughout the body), renal failure, permanent brain injury and even death (Black, Allen, Bhutta,.....et al., 2008; Farneti& Genovese, 2017).

Neonatal dysphagia is common among high risk neonates with risk factors such as premature birth, low birth weight, birth asphyxia, congenital anomalies and other neurological damage categories (Jadcherla, 2016). Commonly reported neonatal dysphagic symptoms in oro-pharyngeal and phryngo-esophgeal phase of swallowing were latching related difficulties,

delayed sucking, lack of rhythmic sucking and tongue movements, reduced extraction of bolus, regurgitation, delayed initiation of pharyngeal phase of swallow, silent aspiration, penetration, failure of peristaltic movements, arching, gagging, irritability, pooling, wet gurgly voice/breathing, coughs while feeding, stridor, disco-ordination in the pharyngo-upper esophagealsphinctric movements, apnea, feeding related bradycardia, desaturations and other cardiorespiratory events (Jadcherla, 2016).

Aspiration can also occur normal subjects. However, it is generally cleared by their body's defence mechanism, making it lower risk for any serious issues (Gleeson, Maxwell & Egli, 2011). Therefore, serious feeding and swallowing issues are rare in healthy typically developing children (Borowitz & Borowitz, 2018).

### **Causes of neonatal dysphagia**

Neonatal dysphagia often occurs as a result of multi-factorial causes. It occurs due to multiple combinations of medical conditions, which could be long term. In some cases, it is temporary due to immaturity or temporary aberrations in the central nervous system which subsides spontaneously. It is always necessary to identify the root cause of neonatal dysphagia to initiate appropriate intervention at the earliest to prevent further complications. Any delay in establishing root cause of the feeding and swallowing difficulties to make appropriate alternatives to main the optimum nutrition can lead to severe complications in the pulmonary system and failure to thrive. Some of the causes of neonatal dysphagia and patterns of associated feeding and swallowing difficulties are given below.

#### **a) Prematurity**

Gestational age plays a key role in the maturation of anatomical and physiological structures especially in preterm babies. The patterns of swallowing in premature infants are

different from those of full-term infants. Lau, Alagugurusamy, Schanler, Smith and Schulman in 2000 reported a significant association between the gestational age and the maturity level of the child's sucking. During premature birth, the development and refining of these oral feeding and swallowing skills in the last trimester is disrupted. This disrupted neuro-typical developmental pattern of oral feeding skills has a long-term impact on oral feeding capabilities of the neonate, nutrition, growth, mother-child bonding, and interaction (Bruns & Thompson, 2012).

Extremely preterm neonates tend to have weak sucking reflex, more oral fatigue and aspirate more, and are not the candidate for oral mode of feeding (full enteral feeding). During this period, they require assistance with respiration, feeding and swallowing. Therefore, intra-gastric tube feeding along with occasional oral feeding is supplemented in preterm neonates. Further, these feeding and swallowing difficulties are temporary in nature and it spontaneously subsides with child's growth and development (Arvedson, 1998).

Preterm neonates usually present with a disorganised suck pattern. This pattern reflects a general immaturity of organisation of behaviour and responses. It is characterised by three to five brief suck bursts with lengthy pauses between the episodes of sucking activity. There is also marked habituation to the teat, so sucking will frequently fail to recommence spontaneously. A new stimulus such as movement of the teat by the feeder is required before further sucking activity is noted. They can also have a dysfunctional suck pattern which demonstrates aberrant rather than immature features. It is suggestive of underlying neurological problems whose other signs may not yet be manifested. Sucking patterns of preterm neonates often remain significantly less co-ordinated and less efficient than those of full-term infants at term age and beyond (Dodrill, 2011). Mathew in 1988 stated that there is a high incidence of apnea events in preterm

infants during oral feeding. Therefore, they were considered as a “high-risk” group for early feeding difficulties.

The extremely preterm neonates along with associated medical conditions such as anoxia or hypoxia can have permanent neurological damage which in turn leads to developmental disorders such as cerebral palsy (CP). Dysphagia is commonly seen in CP children with more deficits in oral phase of swallowing due to inadequate functioning of the oral muscles in maintaining lip seal for mastication and transporting the bolus to the pharyngeal area, impaired palatal functions, lack of salivary control which in turn leads to drooling, hypersensitivity in the oral area (particularly tactile), tongue thrust, prolonged and exaggerated bite reflex and hypo/hyper active gag reflex. They also have associated motor deficits that causes poor head and neck control (proper positing for feeding), poor co-ordination of the muscles and dysfunction in the extremities (more in the upper part of the body) (Wagner, Rudolph, & Singleton, 1968). Full term neonates usually display food seeking behaviours by rooting around the chest for the nipple of breast/bottle, whereas preterm neonates slowly acquire all these skills as they move towards term (Arvedson, 2008).

#### **b) Very low birth weight**

Very low birth weight is commonly seen in preterm births which increases the risk for feeding and swallowing difficulties. Minde, Perrotta, and Marton in 1985 found that neonates with very low birth weight were characterized as being disorganized, with a weak suck, poor lip seal, presence of frequent vomiting and problems with the co-ordination of sucking, breathing and swallowing, and they were also more prone to Gastro-esophageal reflux disease (GERD). In very low birth weight neonates, approximately around 80% even at 6 months corrected age did not develop age-appropriate oral-motor function, eating and drinking skills, diet (including food

consistencies) and behaviours at mealtimes, suggesting significant ongoing developmental compromise (Mathisen, Worrall, O'callaghan, Wall, & Shepherd, 2000).

**c) Congenital defects of the upper airway**

Congenital defects in upper airway passage such as defects in nasal or nasopharyngeal (atresia or stenosis, tumours and infections) and oro-pharyngeal region (cleft lip and or palate, deformities of the lips, tongue and palate) can produce dysphagia. These neonates with congenital upper airway defects tend to accept oral feeds, eagerly suck and swallow, but only for shorter span of time. Eventually, they tend to have a reduced co-ordination in the sucking, swallowing and breathing sequence. This in-coordination leads to penetration, aspiration, choking, and oral fatigue within shorter span of feeding and ultimately to respiratory arrest (Wagner et al., 1968).

**d) Congenital defects in larynx, trachea and esophagus**

The congenital defects in the laryngeal (webs, cysts, clefts, stenosis, vocal cords paralysis in midline position and tumours), tracheal and esophageal region (cleft and fistula in tracheoesophageal region, stenosis or achalsia, atresia, vascular rings, cysts, and strictures) can lead to dysphagia. Particularly they exhibit eagerness for sucking, normal and effective sucking and swallowing with breathing co-ordination. However, after swallowing it leads to aspiration, obstruction in the airway and further it causes pneumonitis. In some structural defects like congenital strictures, dysphagia may not be evident until solid foods are introduced (Wagner et., 1968). In many cases, dysphagia due to congenital defects requires emergency intervention to preserve life.

**e) Inflammatory condition**

There are two types of inflammatory disorders, infectious and non-infectious. If the infectious disease affects the gastrointestinal tract, then it can cause dysphagia or odynophagia (Arvedson, 1998). Further, hypertrophy of the adenoid and tonsillar region can obstruct the airway, which in turn leads to feeding and swallowing difficulties. Even infections in the laryngeal region can lead to sudden onset of dysphagia. The nature, extent and type of dysphagia could vary depending on the aetiology.

Dysphagia manifestation in non-infectious condition (endocrine disorders) such as diabetes, hyperthyroidism, and myxedema can rarely be seen. Children with these may develop symptoms such as gastroesophageal reflux (GERD), esophagitis, delayed gastric and esophageal emptying (Ricciordelli& Richardson, 1991).

**f) Neoplastic conditions**

Dysphagia can be seen in cases of aerodigestive tract's benign and malignant neoplasms. In these cases, liquid foods can be tolerated, but the presence of obstructive lesion causes choking or regurgitation of solid foods. It can also produce stridor, airway distress, and aspirations (Hassall, Dimmick& Magee, 1993).

**g) Central nervous system disease**

Dysphagia is one of the earliest signs of neurological impairments such as microcephaly, hydrocephaly, macrocephaly and so on. The term “neurogenic dysphagia” is used to describe any swallowing impairments due to neurological impairments. In neurogenic dysphagia, weak sucking reflex, aspiration of their own secretions, absent arousal reflex, poor gag reflex and nasal regurgitation can be seen. Along with these characteristics, they tend to have poor neck control

and muscle tone. If this left untreated, it leads to recurrent pneumonia due to aspiration and failure to thrive (Bluestone, 2003).

#### **h) Neuromuscular disease**

Upper gastro-intestinal dysfunctions are common in children with neuromuscular disease due to protein manifestations. As the result, they tend to exhibit dysphagia with symptoms such as choking, frequent throat clearing during or immediately after eating, vomiting, gagging, poorly co-ordinated swallowing movements and aspiration. In some cases like familial dysautonomia, disordered swallowing is observed due to delayed opening of crico-pharyngeal muscles, changes in the esophageal motility with improper lower esophageal sphincter relaxation. The prognoses in these cases are poor (Bluestone, 2003).

#### **Incidence and Prevalence of neonatal dysphagia**

The incidence of feeding and swallowing disorders is assumed to increase due to the improved survival rates of children with complicated medical conditions (Lefton-Greif, 2008; Newman et al., 2001). This could be attributed to advances in medical technology resulting in higher rates of survival. As per the survey interview of Centers for Disease Control and Prevention, 0.9% of children (around 569,000) in the age range of 3 to 17 years reported swallowing difficulties (Bhattacharyya, 2015).

According to the National Hospital Discharge Survey in 2010 (by National Centre for Health Statistics), approximately 116,000 new-borns are discharged from hospitals with the diagnosis of feeding problems. Among children with developmental disorders, the incidence is estimated around 30%-80% (Arvedson, 2008; Lefton-Greif, 2008). Further in children with CP, incidence of oro-pharyngeal dysphagia is estimated to be around 19.2% –99.0%. This values

rise with severe cognitive impairment and decline in gross motor function (Erkin, Culha, Sumru, &Gulsen, 2010).

The exact prevalence of dysphagia in neonates and/or feeding and swallowing issues in neonates is not known. The prevalence of feeding and swallowing related issues in typically developing children is estimated to be around 25% to 45% whereas in children with developmental disorders is around 33% to 80% (Dahl, Thommessen, Rasmussen, &Selberg, 1996; Reilly, Skuse, & Poblete, 1996; Burklow, Phelps, Schultz, McConnell, & Rudolph, 1998; Schwarz, Corredor, Fisher-Medina, Cohen, &Rabinowitz, 2001; Fieldet al., 2003).

Feeding and swallowing problems are frequently reported in preterm and low birth weight infants (Zehetgruber, Boedeker, Kurth, Faas, Zimmer &Heckman, 2014). The prevalence of feeding and swallowing issues in preterm neonates with gestational age between 28 – 37 weeks is approximately around 10.5% and this percentage approximately increases to 24.5% for those who are born with very low birth weight (VLBW) of less than 1.5 kg (Motionet al., 2001).

### **Studies investigating neonatal dysphagia**

Mercado-Deane et al. (2001) assessed the incidence of swallowing dysfunction in infants with vomiting or respiratory symptoms. This was a retrospective study. They found that the incidence of swallowing dysfunction was significant in premature infants and in those with bronchopulmonary dysplasia, congenital heart disease, esophageal atresia or tracheoesophageal fistula, various syndromes and neurological abnormality.13.4% had swallowing dysfunction which included aspiration and penetration.

Jadcherla et al. (2009) aimed to determine pharyngoesophageal motility correlates in neonates with dysphagia. Twenty dysphagic neonates underwent a swallow-integrated



pharyngoesophageal motility assessment of basal and adaptive swallowing reflexes using a micromanometry catheter and pneumohydraulic water perfusion system. They found that pharyngoesophageal manometry correlates were significantly different between the primary oral feeders versus the chronic tube feeders for swallow frequency, swallow propagation, presence of adaptive peristaltic reflexes, oral feeding challenge test results, and upper esophageal sphincter tone. They concluded that the dysfunctional neuromotor mechanisms may be responsible for neonatal dysphagia or its consequences. They also found that 30% had nasopharyngeal reflux, 35% experienced pooling, 35% had delayed swallow, 55% had aspiration, and 90% experienced laryngeal penetration using videofluoroscopy. The video fluoroscopy characteristics were similar between patients with feeding success ( $n = 15$ ) and those with feeding failure ( $n = 5$ ).

Lee, Chang, ..., and Park (2011) detected swallowing dysfunction using modified barium swallow (MBS) and determined risk factors for swallowing dysfunction in very low birth weight (VLBW) infants with oral feeding desaturation near discharge. They retrospectively reviewed 41 VLBW infants referred for MBS test. Infants who showed impaired airway protection, including inadequate epiglottic closure, laryngeal penetration and/or tracheal aspiration by MBS test, were compared to those without impaired airway protection. Eleven infants (26.8%) showed impaired airway protection by MBS test. They had a significantly lower gestational age at birth but a similar postmenstrual age compared to those without impaired airway protection. All infants with impaired airway protection were born at  $\leq 28$  weeks of gestation. They concluded that swallowing dysfunction resulting in aspiration should be considered as a cause of significant oral feeding desaturation in infants born at  $\leq 28$  weeks of gestation regardless of postmenstrual age.

A pilot study by Ferrara, Kamity, Islam,.... and Hanna (2018) determined the pharyngeal swallow in preterm infants with dysphagia. This study included nine preterm

infants who demonstrated clinical symptoms of dysphagia. These babies underwent video fluoroscopic swallow study with thin barium at room temperature using the nipple flow rate they had been using bedside. They found that all nine infants demonstrated pharyngeal phase dysphagia.

Krüger, Kritzinger, and Pottas (2019) aimed to identify symptoms of oropharyngeal dysphagia in breastfeeding neonates with hypoxic-ischemic encephalopathy (HIE) on therapeutic hypothermia (TH). 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 received whole-body TH. Feeding was clinically evaluated by a speech-language pathologist using the Preterm Infant Breastfeeding Behavior Scale. Twenty-five neonates (89.2%) had at least one symptom of oropharyngeal dysphagia. Falling asleep during feeding, noticeable oral secretions, coughing, and flaring nostrils were symptoms of dysphagia 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 new-borns. The HIE group had significantly more closed eyes and minimal movement during breastfeeding, while controls showed the quiet-alert state ideal for breastfeeding.

Han, Shin, and Jeon (2020) determined the prevalence, characteristics, and risk factors of swallowing dysfunction in infants admitted to the neonatal intensive care unit from 2016 to 2018, on whom modified barium swallow study was performed due to oral feeding difficulties. Among a total of 54 infants enrolled, nine (16.7%) were term infants, 13 (24.1%) were late preterm infants (gestational age, 34-36 weeks), and 32 (59.3%) were early preterm infants (gestational weeks). Gestational age and birth weight were smaller in infants with swallowing

dysfunction. Total duration of mechanical ventilation and duration of invasive ventilation were longer in infants with swallowing dysfunction. The risk of swallowing dysfunction increased by 11.2 times for infants with gestational weeks compared to infants with gestational weeks. They concluded that preterm infants with gestational weeks or with longer ventilation duration are at a higher risk of aspiration.

### **Significance of early identification of feeding and swallowing problems**

Sucking and swallowing abnormalities in early infancy have long been viewed as potential markers of neonatal brain injury. It could be the first indication that the infant has neurological problems (Reilly & Skuse, 1992). Moreover, it has been suggested that prolonged dysphagia or swallowing difficulties in very preterm infants may represent an early marker of undiagnosed brain injury.

Successful feeding depends upon the co-ordination of sucking, swallowing and breathing (Arvedson & Brodsky, 2002). Neurological controls that support this co-ordinated motor activity are complex and rely upon the integration of cortical, sub-cortical, brainstem and cerebellar inputs (Barlow & Estep, 2006). It is generally agreed that neonatal brain injury can damage these neural pathways with resultant sucking and swallowing problems (Parkes, Hill, Platt, & Donnelly, 2010).

In addition, it is argued that a relation exist between early sucking and swallowing difficulties and later neurodevelopment. In a recent review, Poore and Barlow (2009) contend that early sucking skills may predict later neuro-developmental outcomes. Particularly, there are potential links between the neural networks that control sucking, swallowing and speech.

Therefore, they referred them as early predictors of feeding and swallowing related issues which in turn helps in early intervention.

Slattery, Morgan, and Douglas in 2012 conducted a systematic review of literature on early sucking and swallowing problems as predictors of neuro-developmental outcome in children with neonatal brain injury. The objective of their study was to describe the relation between early measures of sucking and swallowing and neuro-developmental outcomes in neonates diagnosed with brain injury and infants born very preterm (<32 weeks) with very low birth weight (<1500g), at risk of neonatal brain injury. The authors found early sucking and swallowing problems in consistent proportion of infants (35-48%) with varied aetiologies of neonatal brain injury. But the available evidence was insufficient to establish the exact relation between neonatal brain injury and early sucking problems. Further, the authors indicated that more detailed research is required to delineate the later neuro-developmental outcomes based on the early measures of sucking and swallowing functions.

Early detection of dysphagia in their neonatal stage itself is very important to prevent or minimize the complications as well as the negative impact of feeding and swallowing difficulties on the child as well as on the caregiver. Early detection also enables the health care providers to create awareness among the parents/ caregivers about signs and symptoms of pediatric dysphagia as well as the need to approach respective health care professional to receive appropriate management. If those high-risk children are left untreated, then it can lead to failure to thrive, inability to maintain adequate hydration and nutrition, gastroesophageal reflux (GERD), aspiration pneumonias and so on (Prasse&Kikano, 2009).

## **Evaluation of feeding and swallowing in neonates**

Additionally, the clinical evaluation of the neonates at risk of having feeding and swallowing related issues is the vital part of the evidence-based practice in the field of neonatal dysphagia (Thoyre et al., 2013). The main objective of the detailed clinical evaluation of feeding and swallowing is to establish and understand the nature of the problem, to know about the parental perception of the problem, to identify the neonates' oral readiness for feeding, to make an appropriate differential diagnosis as well as to determine the individualized intervention plan (Arvedson, 2008 &Thoyre et al., 2013).

Clinical feeding assessment includes both subjective assessment (includes non-instrumental assessment such as scales/checklist/test materials) and objective assessment (includes instrumental assessment such as Modified Barium Swallow Study). The subjective assessment of feeding and swallowing refers to the direct evaluation of feeding swallowing and its associated behaviour in the clinical population without the use of instrumentation. Though the instrumental assessment provides us with objective data, the importance of the subjective assessment cannot be undermined. There are three main components of the assessment; parental interview and the review of the medical chart to obtain information on birth, medical, feeding and developmental history along with detailed clinical feeding and swallowing assessment to correlate and support the diagnosis (Arvedson, 2008; Lau & Smith, 2011). The use of validated instruments as a part of the subjective assessment is essential in clinical practice, because it provides a common language among clinicians, facilitates the production of diagnostic data and promotes the evaluation of techniques and approaches used during clinical assessment (Brandao, Dos Santos, &Lanzilotti, 2013).Table 2.1 provides information on different components of subjective feeding and swallowing assessments and its significance.

Table 2.1

*Various components of subjective feeding and swallowing assessments and their significance*

<b>S.No</b>	<b>Components</b>	<b>Significance</b>
1	Physiological functioning of system	Airway stability is a prerequisite for effective and successful feeding. The most common causes of neonatal dysphagia are respiratory problems. Possible chronic aspiration and other signs of dysphagia may also be identified by assessing the respiratory rate. Therefore, it is important to evaluate respiratory patterns during feeding sessions.
2	State of alertness during feeding sessions	The neonate should be in optimum alertness for an efficient and successful feeding. Because the state of neonates usually varies during feeding, this should be measured in order to determine the optimal alertness for proceeding with oral mode of feeding.
3.	Stress cues during feeding sessions	In feeding readiness, the capacity of neonate to react to incoming sensory information's plays a significant role. The relationship between state regulation of the neonate, motor and autonomic nervous system should be examined in order to determine any sort of stress cues during feeding sessions, if any, to enable the health care professional or parent to make necessary actions.

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4	General screening of muscle tone and movements	For efficient and safe feeding, appropriate postural control is a necessary. Inadequate tone of muscles and reduced postural control can adversely affect the oral mode of feeding. When issues are found with muscle tone and postural control, referral may be made to an occupational therapist and/or physiotherapist.
5	Oral peripheral mechanism examination	Successful and safe swallowing requires functions of 31 muscles and five cranial nerves to be synchronized. Anatomy, physiology, primitive oral reflexes and cranial nerves underlying the swallowing functions of the neonates should be assessed
6	Nature of feeding and swallowing problem	The aim of the clinical assessment is to observe the oral preparatory phase as well as the oral transport phase of swallowing to draw inferences about the pharyngeal phase of swallowing, also to provide specific information on management strategies and to track the progress.
7	Mother-child interaction during feeding	The neonate with risk factor has heightened potential to develop difficulties in interactions with parent/caregiver during feeding. Success in feeding the neonate depends on the ability of the parent / caregiver to track the stress cues of the neonates if any and to make necessary environmental modifications to promote successful performance in oral feeding. It should be noted that

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parent-infant interactions during the sessions of feeding provides a basis for social interactions as well as reciprocity of the communication interaction.

- 8 Use of compensatory strategies in feeding sessions
- The health care professionals involved in dealing with neonatal population (particularly neonatologist, nurses, SLP) should be able to suggest compensatory approaches as part of the initial evaluation to facilitate effective feeding in the neonates. Strategies to be considered may include modifications in the neonates position during breast or bottle feeding, nipple style used (nipple slit or other modification) or external alterations in the feeding pace. These approaches will allow the mother to feel the control in the feeding sessions, which in turn will help her to build confidence in meeting the nutritional needs of her child.

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Screening using a validated material is an important part of clinical evaluation and serves as a first step for the early identification of feeding and swallowing difficulties in neonates (Thoyre, Park, Pados, & Hubbard, 2013). Screening tools for dysphagia are widely used to determine the presence or absence of dysphagia. Further, questionnaires are widely adopted method for screening tools among the various other methods to classify and characterize the signs and symptoms of any condition including dysphagia (Orenstein, 2006). This is because the professionals can quickly administer the questionnaires on the clinical population without any rigorous training. Moreover, in many health care setups, instruments may not be available



due to various reasons. The use of these instruments can be invasive and may need physical handling, which can lead to discomfort for the neonate. Studies by Philbin and Ross (2011) as well as Browne and Ross (2011) indicated that unnecessary physical handling may disrupt state regulation during this sensitive stage of neurological development. In addition, the main purpose of the screening tool for dysphagia is to identify presence of dysphagia as early as possible to make referral for detailed evaluation and appropriate intervention plan (Leder&Espinosa, 2002; Duarte, 2010). Some of the screening tools have been discussed below.

Dysphagia Screening Test for pre-term infants (DST-PI, Lee & Seo, 2017) is a valid and reliable tool for the detection of dysphagia particularly in pre-term neonates. This screening test consists of seven test items such as gestational age, history of apnea, cyanosis during feeding, decreased oxygen saturation after feeding but within three minutes, coughs during or after feeding, voice change after feeding and swallowing patterns. Further, sensitivity and specificity of DST-PI was established for the detection of presence of penetration (at supra-glottic level) and aspiration (at sub-glottic level) in pre-term infants.

Feeding and Swallowing Scale for Premature Infants (FSSPI, Moon, Jung, Cheon, Oh, & Kwon, 2017) was developed with reference to Baby Regulated Organization of Subsystems and Sucking approach (BROSS) approach which consists of 6 consecutive developmental feeding levels such as non-nutritive, obligatory, alternating, intermittent, co-ordinated and integrated phases. The FSSPI consists of the following six items such as non-nutritive sucking, nutritive sucking, sucking pattern, sucking burst, developmental stage and regularity of rhythm. Also, it has been verified for its reliability and validity in premature infants with gestational and corrected age of less than 34 weeks and 3 months respectively based on Video Fluoroscopic Swallowing Study (VFSS). The results showed a high degree of both intra-rater reliability and

inter-rater reliability. However, there was a significant negative correlation between the FSSPI score and corrected age at the time of performing VFSS.

Another screening tool titled “Infant and Child Feeding Questionnaire” (ICFQ) was developed to facilitate early detection of feeding and swallowing problems (Byrd, Steinfeld, Hoffmann, & Silverman, 2017). This early detection is achieved by promoting effective communication between caregivers and health care providers, resulting in referral for evaluation and treatment of feeding and swallowing problems to the specialists. The authors also determined that whether items from the ICFQ could be used to look for differences between children with known feeding problems (FP) and without known feeding problems (NFP). Caregivers of children ages 36 months or younger with FP and NFP were recruited to complete the ICFQ as well as demographic questions. 64 caregivers of children with FP and 57 caregivers of NFP children were recruited. Based on the caregiver’s response of both the groups, they identified 9 feeding behaviours that distinguished between the groups. Of these, a combination of 3 questions in ICFQ was seen in FP than in NFP group. Overall results revealed that the subset of items in the ICFQ has promise for distinguishing FP from NFP groups. However, this tool has not been validated.

There are also a few tools that assess only the breast-feeding behaviours in infants. The Infant Breast-Feeding Assessment Tool (IBFAT, Matthews, 1988) is one such tool that was designed to measure four major components of infant breastfeeding behaviors. The four components were readiness to feed, rooting, fixing, and sucking. The scoring range for each component is 0 to 3, giving a total score ranging from 0 to 12, with 12 being the score for vigorous, effective feeding. In addition to the four components above, two non-scoring items

describing infant' s state and mother' s satisfaction with the breastfeeding experience are included in the tool.

The Systematic Assessment of the Infant at Breast (SAIB, Shrago&Bocar, 1990) is another tool designed to assess the infant's "contribution to breastfeeding"; that is, whether the infant is able to extract milk from the breast effectively, thereby contributing to successful breastfeeding. The tool consists of observations in the categories of alignment, areolar grasp, areolar compression, and audible swallowing. The authors state that the SAIB can serve as a systematic teaching guide and can be administered by professionals and mothers; however, they fail to provide a scoring system.

The Mother-Baby Assessment (MBA, Mulford, 1992) is designed to assess five sequential breastfeeding behaviors of both mother and infant, based on the assumption that breastfeeding is a mutual effort. The five steps of breastfeeding are signalling, positioning, fixing, milk transfer, and ending. Ten is the highest possible score: A 5 for maternal behaviors and a 5 for infant's behaviors in each of the steps indicate highly effective feeding.

## **infant's SAIB is designed to assess the**

The LATCH (Jensen, Wallace &Kelsay, 1994) was designed to identify areas of needed intervention and to determine priorities in providing patient care and teaching. The acronym "LATCH" represents the five components of assessment. They are the infant' s ability to "Latch" correctly onto the breast, the amount of "Audible" swallowing noted at breast, the mother' s "Type" of nipple, the mother' s "Comfort" level regarding her breast and nipples, and the amount of "Help" the mother needs to hold her baby to the breast. The scoring range for each component is 0 to 2, for a possible total of 0 to 10 points.

The Pre-term Infant Breastfeeding Behavior Scale (PIBBS, Nyqvist, Rubertsson, Ewald, & Sjoden, 1996) is designed to assess breastfeeding competence in preterm infants by both professionals and mothers. The tool consists of operational definitions of maturational steps in six behaviors: rooting, areolar grasp, longest duration of latching on, sucking, longest sucking burst, and swallowing. Behaviors are rated with different ranges (0-2, 0-3, 0-4, and 0-6), giving a total score ranging from 0 to 20.

In addition to the above tools, a look into the existing literature revealed that there are several in-depth assessment tools for feeding and swallowing related issues in the children. Some of the published in-depth feeding assessment scales and checklists which can be useful in systematizing the observation have been described below.

The Neonatal Oral Motor Assessment Scale (NOMAS) given by Palmer, Crawley and Blanco (1986) is an evaluation of oral motor patterns during nutritive and non-nutritive sucking in infants up to 3 months of age that discriminates normal from abnormal suck and quantifies the degree of abnormality. It primarily considers the tongue and jaw movements during sucking and classifies them into normal, disorganised and dysfunctional. Revised NOMAS identifies different oro-motor components as efficient and inefficient feeder on a sample of high risk premature neonates (Palmer et al., 1993). Further in NOMAS, the developers did not assess its inter-rater reliability at the time publishing their final scale. Later, Van der Schans and Da Costa in 2008 reported a moderate to substantial agreement on evaluating the inter-rater reliability of NOMAS. Further, the validity of the NOMAS needs to be established in future research (Howe, Sheu, Hsieh, & Hsieh, 2007).

Developmental pre-feeding checklist (Morris & Klein, 1987) is a diagnostic tool developed for assessing the developmental skills emerging between 0 to 24 months of age. It provides both qualitative and quantitative description of feeding performance, brief history of feeding problems and also detects any abnormal oral pattern, if present. It assesses 15 skills with reference to its approximate age of emergence such as feeding position (1 month to 18 months), food quantity (1 month to 18 months), food types eaten (birth to 7 months), sucking liquids from the bottle or breast (1 month to 12 months), sucking liquids from the cup (4 months to 24+ months), sucking soft solid or pureed food from the spoon (under 3 months to 24+ months), swallowing liquids (1 month to 24+ months), swallowing semisolids (under 3 months to 24+ months), swallowing solids (6 months to 24+ months), co-ordination of sucking, swallowing and breathing (1 month to 15 months), control of drooling (1 month to 24 months), jaw movements in biting (5/6 months to 24+ months), jaw movements in chewing (under 5 months to 24+ months), tongue movements in chewing (under 6 months to 24+ months) and lip movements in chewing (under 6 months to 24 months). This scale provides systematic observations through insight on normal oral sensori-motor development along with feeding functions, but it lacks interpretation guidelines. However, this tool is not standardized for feeding assessment.

The Clinical feeding evaluation of infant's scale (Wolf & Glass, 1992) is a validated scale to assess the feeding difficulties among infants in the age range of 0 to 2 years who are fed by bottle or breast, also assesses difficulty which may be present in spoon feeding as well as in cup drinking. It provides a comprehensive feeding performance assessment for high risk babies in neonatal intensive care unit. It assesses the infants' feeding function using traditional approaches of oral sensory responses and motor control along with evaluation of other additional factors that contribute to the feeding function. It primarily focuses on areas of state and

behaviour, oral motor function, motoric control, physiologic control, tactile responses and co-ordination of sucking, swallowing, and breathing. Evaluation is done based on the information from medical tests and procedures along with skilled clinical observations.

Infant feeding evaluation (Swigert, 1998) is a non-standardised evaluation and used in documenting a variety of observations including infant's responses to attempted interventions. It can be used in the age range of 0-4 months but certain components of this scale are specified only for preterm or ill infants.

Early Feeding Skills Assessment for preterm infants (EFS, Thoyre, Shaker, &Pridham, 2004) is a checklist for assessing infant readiness and tolerance for feeding and to profile the infant's developmental stage according to their feeding skills, that is, the abilities to remain engaged in feeding, organized oral motor functioning, coordinate swallowing with breathing and maintain physiologic stability. However, no data was provided regarding content validity, inter and intra-rater reliability.

Neonatal Feeding Assessment Scale (NFAS, Viviers, Kritizinger, &Vinck, 2016) is a clinical feeding assessment scale for very young neonates in South Africa. The components consist of physiological functioning, state of alertness during feeding, stress cues during feeding, general movements and muscle tone screening, oral peripheral examination and clinical feeding and swallowing evaluation (nutritive and non-nutritive sucking). The tool is lengthy and time consuming for clinical examination. Also, the reliability, validity and psychometric properties of this scale were established, which that shows that NFAS is a promising tool for the early identification of oro-pharyngeal dysphagia.

Even though, there are several assessment scales and checklists for the identification of dysphagia, screening tools to identify neonatal dysphagia, caused due to different etiologies and irrespective of the way feeding is done (breast/bottle/spoon/any other mode) are limited. Although the recognition of the unique needs posed by neonatal dysphagia has been increased, research into the development of clinical assessment tools, particularly the screening tools for this population continues to remain limited (Heckathorn, et al., 2016 & Jadcherla, 2016). Further, there are no validated tools that cut across the underlying medical condition and specify the critical test items along with well-established scoring and interpretation algorithms for assessing neonatal feeding and swallowing difficulties.

Further in 2003, the World Health Organization (WHO) and UNICEF adopted the “Global strategy for infant and young child feeding” and identified that different feeding practices exist across worldwide from birth to infancy which is related to their culture, socio-economic status and educational backgrounds, therefore regional specific practices should be given importance during evaluation and management of feeding disorders in infants. Thus, there is a need to develop a context specific, validated screening tool to assess dysphagia in neonates, which would further facilitate management. This would facilitate the speech pathologists to evaluate these infants and make informed decisions on the appropriate management of dysphagia in high risk neonates.

## **CHAPTER III**

### **Methods**

The present study aimed at developing and standardizing a screening tool for the early identification of dysphagia in the neonatal population. The current study was executed in four different phases which were as follows:

Phase I - Construction of the Neonatal Dysphagia Screening Tool (NDST)

Phase II- Clinical validation of NDST

Phase III – Assessment of sensitivity and specificity of NDST

Phase IV- Assessment of inter-rater reliability

#### **PHASE I: Construction of the Neonatal Dysphagia Screening Tool (NDST)**

The following steps were undertaken in the current research as part of the NDST construction.

##### **Step 1: Development of the preliminary version of the NDST**

This step involved the development of a preliminary version of the NDST which was in the form of a questionnaire to identify the presence of feeding and swallowing difficulties if any, in the high-risk neonates. The process of development of NDST commenced with an extensive review of the available literature on neonatal dysphagia and its common aetiologies particularly in the Indian context. Then it progressed to reviewing of normal feeding and swallowing milestones, various signs and symptoms of dysphagia, challenges faced in feeding neonates such as feeding positions, feeding intolerance and so on. It also involved reviewing the published feeding and swallowing assessment scales and checklists in order to identify and select the



appropriate content. In particular, the questions were selected based on the aspects that can be observed by the mothers during their feeding sessions. Appropriately selected content and test items were organised in the order of before feeding, during feeding and after feeding.

The preliminary version of the NDST consisted of five sections which included demographic data, medical history, physical/physiological functioning, feeding history and feeding and swallowing assessment. The demographic data section consisted of information such as gender, chronological, gestational and corrected age, socio-economic scale (SES) and other basic contact details. The medical history section of NDST consisted of sections pertaining to pre-natal, peri-natal and post-natal history of the child. The physical/physiological functioning section consisted of information pertaining to neonatal state, oral reflex and oro-motor examination. The feeding history section of NDST consisted of questions to elicit information about the feeding method, use of feeding aids, feeding duration, feeding frequency, perception of feeding cues and maternal satisfaction with the feeding process. The feeding and swallowing screening section of NDST comprised of 18 questions related to feeding and swallowing difficulties frequently seen in neonates which were organized in the order of before (3 questions), during (5 questions) and after (10 questions) feeding.

### ***Scoring system of NDST***

For NDST, binary yes/no scoring system was chosen to record the mother's responses. In this binary yes/no scoring system, "yes" indicated the presence of difficulties and "no" indicated the absence of difficulties. In order to get an objective score from the obtained data using NDST, a 'yes' response was assigned a value of "1" and a 'no' response was assigned a value of "0".

## Step 2: Content validation of NDST

### *Preparation for content validation*

The content validity of the preliminary version of the NDST (18 questions) and its rating scale was assessed by obtaining feedback from a multi-disciplinary expert group of professionals. A feedback protocol was designed with a 3-point rating scale (2 indicating appropriate, 1 indicating nearly appropriate and 0 indicating inappropriate) to judge the appropriateness of the 18 questions along with some allocated space for suggestions under each question. Participants were provided with written instructions for rating the questions in the NDST using the scale. Figure 3.1 depicts a part of the feedback protocol used for the content validation of NSDT.

**Feeding and swallowing assessment**

**\*Note:** Rate each questions as 0- Inappropriate, 1- Nearly appropriate and 2-Appropriate. If scored 0/1, Please indicate the reason for the same and suggest an alternative.

S.No	Questions	Appropriateness
1.	Does your child have difficulty in turning his/her body towards you during feeding? Yes / No	2- Appropriate 1- Nearly appropriate 0- Inappropriate
<b>Suggestions:</b>		

Figure 3.1. Screenshot of the feedback protocol used for content validation of NSDT.

### ***Details of the content validators***

Ten multi-disciplinary experts were recruited as content validators to establish the content validity of the NDST. Amongst them, seven members were experienced speech-language pathologists (SLP's) in the field of paediatric dysphagia and other three members were medical professionals such as neonatologist and neonatal nurses who had an expertise in the field of neonatology. Table 3.1 depicts the details of the content validators.

### ***Participant selection criteria for content validation***

The participants who fulfilled the following criteria were considered for inclusion for content validation:

- Participants with a minimum professional qualification of master's degree in their respective fields.
- Participants with a minimum of two years of clinical experience in the field of neonatal/paediatric dysphagia or feeding and swallowing issues.
- Participants who resided and worked in India to ensure the relevancy of their clinical experience to the Indian context.

Table 3.1:

*Details of the content validators*

<b>Details of the content validators</b>	<b>Number of participants</b>
<b>Professionals</b>	
Speech-Language Pathologist	7
Neonatologist	2
Neonatal nurse	1
<b>Working experience (in years)</b>	
2 -3	5
4-6	2
7-10	1
11-20	2

***Procedure for establishing content validity***

All the participants included for the content validation were orally informed about the nature, purpose and procedure of the study. They were also informed about the instructions and time frame (two weeks) for the completion of the questionnaire given to them. This questionnaire provided the participants with the opportunity to include/exclude/modify any of the questions in the NDST along with commenting on the scoring method used.

***Modifications made in NDST based on the input received***

After the completion of the designated duration, the completed questionnaires were taken back from all the validators. The responses of the validators to the preliminary version of NDST

were noted and appropriate modifications were made. The question “do you feel your child has feeding and swallowing difficulties?” was added because majority of the validators (six validators) recommended to add it. Therefore, it was added in the NDST.

The appropriateness of each question was also obtained from the validators using a 3-point rating scale (score of “2” - appropriate; “1” - as nearly appropriate and score of “0” - not appropriate). Questions which received a score of “2” by more than 80% of the judges were retained. All questions exceeded 80% of appropriateness with the exception of Q16 (Does your child have tiredness/breathlessness and sweating after feeding / falls asleep before the end of feeding?), which had a lower rating for appropriateness. Four validators stated that it would be difficult for the mothers to judge this aspect in the first few days of birth. The others stated it as inappropriate for neonatal feeding. Consequently, Q16 was excluded from NDST. The remaining 17 questions were retained along with a binary choice scoring system. Further, no questions of NDST were modified or rephrased after content validation.

### **Step 3: Pilot study**

A pilot study was carried out on 10 neonates (with and without risk factors) at government hospitals in Mysuru using the content validated NDST. This preliminary pilot study was completed by reviewing the medical records and screening of feeding and swallowing using NDST through an interview with mothers and the responses were documented. Upon completion, the responses were analysed to identify the need for inclusion / exclusion / modification of the items on the content validated NDST based on receptive ability of the parents / caregivers and practical limitations in the hospitals. After the pilot study, it was found that all the test items of the NDST were appropriate and comprehensible by the mothers. Therefore, all

the test items of NDST were retained and finalized for the main study after content validation. It was also seen that the time taken to administer this tool was 10 minutes.

#### **Step 4: Finalization of the NDST**

The finalized version of the NDST was prepared after the content validation and the pilot study. The feeding and swallowing screening section of NDST finally comprised of consisted of 17 questions. The final version of Neonatal Dysphagia Screening Tool (NDST) has been provided in the appendix A.

#### **PHASE II: Clinical validation of NDST**

The finalized version of NDST was administered on the neonatal population to estimate its clinical validity. The details of the participants have been given below:

##### ***Participant details***

A total of 178 mothers of neonates (99 males and 79 females) in the age range of 1 to 5 days born between February and May 2019 were recruited from both private and government hospitals (almost equal number) in and around Mysuru. These participants were divided into three groups based on their medical history. The first group included 75 mothers of neonates (41 males and 34 females) with no known risk factors and this group was labelled as typical group (TG). The second group included 67 mothers of neonates (38 males and 29 females) with high risk factors such as thyroidism, epilepsy and chicken pox (viral/bacterial infections) in the pre-natal period, foetal distress, aspiration of amniotic fluid, moderate to late prematurity (gestational age: 32-37 weeks), low birth weight (LBW-1500 to 2500 g), high birth weight (HBW->4000g), delayed birth cry and moderate birth asphyxia (APGAR scores:4-6) in the peri-natal period and neonatal jaundice, hypoglycaemia (low sugar level), viral/bacterial infections, and high fever in

the post-natal period and this group was labelled as high risk group (HRG). The third group included 36 mothers of neonates (20 males and 16 females) with very high risk factors such as extremely low birth weight (ELBW-<1500g), extreme prematurity (gestational age: 28-32 weeks), serious birth asphyxia (APGAR scores: 0-3), congenital anomalies, respiratory distress syndrome (RDS), hypoxic-ischemic encephalopathy (HIE) and convulsion and this group was labelled as very high risk group (VHRG). The participants were classified into these groups based on the studies by Hawdon, Beauregard, Slattery and Kennedy (2000), Jadcherla (2016), and Farneti and Genovese (2017). The distribution of neonates into the different groups based on their medical history has been depicted in table 3.2.

Table 3.2:

*Distribution of neonates into the different groups based on their medical history*

<b>Medical history</b>	<b>HRG</b>	<b>VHRG</b>
<b>Prenatal risk factors</b>		
Thyroidism	12	0
Chicken pox	1	0
Seizures	1	0
<b>Gestational age</b>		
28-32 weeks (extremely preterm)	0	18
32-37 weeks (moderate to late preterm)	22	0
<b>Birth weight</b>		
<999g (extremely low)	0	3
1000-1499g (very low)	0	6

1500-2499g (low)	17	0
>4000g (high)	1	0
<b>Birth asphyxia</b> (based on APGAR scores)		
0-3 (serious asphyxia)	0	4
4-6 (moderate asphyxia)	13	0
Foetal distress	17	0
Aspiration of amniotic fluid	11	0
<b>Postnatal risk factors</b>		
Congenital anomalies	0	7
Neonatal jaundice	23	0
RDS	0	18
Hypoglycaemia	3	0
Convulsions	0	5
HIE	0	6
High fever & viral/bacterial infection	2	0

Note\*: TG-Typical group; HRG-High risk group; VHRG-Very high-risk group; APGAR- Appearance, Pulse, Grimace, Activity, and Respiration; RDS- Respiratory Distress Syndrome; HIE- Hypoxic Ischemic Encephalopathy. P.S: Some neonates had more than one high risk factor

**Participant selection criteria:** Mothers  $\geq 18$  years of age with atleast 24 hours of feeding experience with their new-borns, who understand and speak Kannada and whose infants were less than 5 days of age and medically stable were included as participants in the study.

All the participants belonged to the socio-economic status scale (SES) II to IV based on the Modified Kuppuswamy scale (Saleem, 2018). The study was carried out by adhering to appropriate ethical procedures. All the caregivers were explained about the purpose of the study



and the procedure involved. A verbal and written consent was obtained from the caregivers. The gender distribution, chronological age and SES of the three groups of participants have been depicted in the table 3.3.

Table 3.3:

*Distribution of participants across the three groups based on the demographic details*

<b>Demographic Details</b>	<b>Groups</b>			<b>Total (N = 178)</b>
	<b>TG (N = 75)</b>	<b>HRG (N = 67)</b>	<b>VHRG (N = 36)</b>	
<b>Gender</b>				
Male	36	39	24	99
Female	39	28	12	79
<b>Chronological Age</b>				
1 day	15	11	8	34
2 days	30	11	8	49
3 days	16	24	9	49
4 days	7	10	4	21
5 days	7	11	7	25
<b>SES</b>				
I	1	0	0	1
II	8	18	5	31
III	30	28	17	75
IV	36	21	14	71
V	0	0	0	0

Note\*: TG-Typical group; HRG-High risk group; VHRG-Very high-risk group;  
SES – Socio-economic status.

### ***Procedure for estimating clinical validity of NDST***

Permission was obtained from the medical superintendent of all the hospitals (both private and government hospitals) where the study was conducted. The procedure for estimating the clinical validity of the NDST began after obtaining an oral informed consent from all the mothers of the participants.

Interview with mothers was conducted along with her description of the feeding and swallowing session, especially with regard to difficulties with feeding and swallowing if any. Medical records were also reviewed in order to obtain the other relevant information. The physical / physiological functioning was tested and the information on history of feeding was also obtained. Following this, feeding and swallowing was assessed using the newly developed NDST. Each question was explained to the mothers of the neonates who were instructed to provide appropriate answers based on their experience of feeding. Information and answers given by the mothers of the respective participant were collected and documented by the researcher.

### **Second assessment using NDST**

All the participants in the three groups, irrespective of presence or absence of feeding and swallowing difficulties, were followed up through the telephonic mode at the end of one month. In the telephone follow up, the relevant medical, feeding and swallowing histories were collected along with the administration of feeding and swallowing questionnaire. These responses of the mothers were documented.

### **Third assessment using NDST**

The third assessment was performed at the end of four months only for the HRG and VHRG. It was planned to carry out this assessment through a face to face parental interview. The TG participants were not included in the third assessment, as they did not exhibit any major feeding and swallowing difficulties. However, this assessment could not be performed on all 103 participants. Only 18 participants reported (14 from HRG and 4 from VHRG) for a face to face interview. The other participants who did not report were assessed through the telephonic mode. However, only mothers of 39 participants could be reached through phone (26 from HRG and 13 from VHRG). The other participants could not be reached due to the change in the contact number or lack of connectivity. Thus, a total of 57 out of 103 participants could be evaluated for a third time on NDST (40 from HRG and 17 from VHRG). Additionally, a screening checklist titled “Remember & Care” (developed at the Department of Prevention of Communication Disorders, AIISH) which screens for motor, hearing, speech and language milestones was administered to assess, if any of the infants exhibited any developmental delays.

### **Phase III- Assessment of sensitivity and specificity of NDST**

In order to assess the sensitivity and specificity of NDST, this was administered on another clinical group of 30 children (20 males and 10 females) with oro-pharyngeal dysphagia in the age range of 0.6 to 4 years. All the participants in the clinical group were recruited from the Department of Clinical Services of All India Institute of Speech and Hearing. Amongst them 21 were diagnosed as Developmental Delay and 9 were diagnosed as cerebral palsy associated with oro-pharyngeal dysphagia by a team of professionals including Speech-Language Pathologist (SLP), Clinical Psychologist, Paediatrician, Physiotherapist and an Occupational Therapist. The

NDST was administered and the feeding problems these children had exhibited at birth and within the first one month of age were documented.

An age and gender matched typical group of 30 children (16 males and 14 females) without feeding and swallowing issues were also included from the nearby play schools. It was ensured that they had no neurological, sensori-motor, communication and cognition related issues based on an informal screening. Further, it was ensured that they had no high-risk factors in their birth/medical history. The NDST was administered on the mothers of this group and the responses were documented.

#### **Phase IV- Assessment of inter-rater reliability**

The NDST was administered on 10% (18) of the mothers of young neonates in the age range of 1 to 5 days by two other qualified SLP's for the purpose of analysing its inter-rater reliability. Before the initiation of data collection, oral instructions was provided to the SLP's about the content, sections, administration procedure and scoring method of this tool. Both the SLP's were blinded to the child and each other's clinical feeding and swallowing assessment.

**Analysis:** The response of the mothers of the neonates on each question was converted into a score based on the rating scale. These were added up to obtain a total score. This was done on the first, second and third administration of NDST. The total score was also obtained from the clinical and control group which were additionally included in the study. The total scores were averaged and obtained for all the groups.

#### **Statistical analysis**

The obtained data was analysed using SPSS (version 20) statistical software. Descriptive statistics was used to obtain mean, median and standard deviation of overall scores obtained on

the NDST. Mann Whitney U test was used to check for any significant differences between the TG, HRG and VHRG that existed for the first (within 5 days of birth) and second administration (at the end of one month) of NDST. Kruskal-Wallis test was used to test the presence of any significant difference across all the three groups (TG, HRG and VHRG) based on the total score for all the three administration of NDST. Wilcoxon test was performed to identify any significant differences between first (within 5 days of birth) and second (at the end of one month) administration of NDST across the groups.

Chi-square test was performed to measure the level of significant association between mother's response and the groups on the NDST. For the inter-rater reliability, Cohen's kappa coefficient was used to identify the level of agreement. Interpretation of the kappa values were done according to the guidelines given by Parikh, Mathai, Parikh, Chandra Sekhar and Thomas (2008).

For the data obtained from the additional clinical and control group, descriptive statistics was performed to obtain mean, median and standard deviation of the overall score. Furthermore, Mann Whitney U test was used to check any significant differences between the clinical and the control groups. Sensitivity and specificity of the NDST were calculated using obtained data in this phase with the formula  $A / (A+C) * 100$  and  $D / (D+B) * 100$  where 'A' is true positive, 'B' is false positive, 'C' is false negative and 'D' is true negative. The results have been presented and discussed in the next chapter.

## CHAPTER IV

### Results and Discussion

The current study aimed at developing a screening tool for the detection of dysphagia in neonates and standardizing it by establishing the validity (content and clinical validity) and reliability (inter-rater reliability). The tool that was developed was referred to as Neonatal Dysphagia Screening Tool (NDST) by the investigators of the current study. The specific objectives of the study were to compare the responses obtained using the NDST across typical, high risk and very high-risk neonates in the age range of 1 – 5 days, to establish the sensitivity and specificity of the developed tool by administering it retrospectively on a known paediatric population with and without dysphagia in the age range of 0.6 to 4 years, to identify the critical test items of the developed tool and to establish its scoring and interpretation algorithm.

The NDST was validated for its contents by a multidisciplinary group of professionals and was subjected to a pilot study, following which the final version was frozen. This final version consisted of five sections which included demographic data, medical history, physical/physiological functioning, feeding history and feeding and swallowing assessment. The NDST was administered on 178 mothers of neonates in the age range of 1-5 days (99 males and 79 females), who were divided into three groups based on the medical / birth history such as typical group (TG-75 participants), high risk group (HRG-67 participants) and very high risk group (VHRG-36 participants), as a part of assessment of clinical validity. The risk factors in the HRG included thyroidism, epilepsy and chicken pox in the pre-natal period, foetal distress, aspiration of amniotic fluid, low birth weight (LBW-1500 to 2500 g), high birth weight (HBW->4000g), delayed birth cry and birth asphyxia in the peri-natal period and neonatal jaundice, hypoglycaemia (low sugar level), viral/bacterial infections, and high fever in the post- natal

period. The VHRG had very high-risk factors such as extremely low birth weight (ELBW- <1500g), prematurity, congenital anomalies, respiratory distress syndrome (RDS), hypoxic-ischemic encephalopathy (HIE) and convulsions. These findings have been presented and discussed under different sections below.

## **I Physiological/physical functioning**

The section on physiological/physical functioning of the NDST included the assessment of neonatal state, oral reflexes, oro-motor structure and functioning. With respect to the neonatal state, it was found that all the neonates of TG and HRG were calm and alert, while 6.1% of neonates in the VHRG were lethargic. These neonates in the VHRG were not aroused by any stimulus and were not moving much as reported by their mothers. This could be attributed to the serious medical conditions present in these neonates. On reviewing the medical history of these lethargic neonates in the VHRG, it was found that all of them had a history of convulsions and were on medication. Eilers and Harrington (2017) reported that neonatal lethargy and convulsions were commonly encountered difficulties for new-borns in the neonatal intensive care unit (NICU). Further they added that neonatal lethargy presents with feeding and swallowing difficulties along with a very poor sucking reflex. Farneti and Genovese (2017) also reported the presence of lethargy while feeding in the new-borns with risk factors for dysphagia. Krüger, Kritzinger, and Pottas (2019) also found that 89.2% of the infants with hypoxic-ischemic encephalopathy with oropharyngeal dysphagia displayed limited arousal during breastfeeding and had significantly more closed eyes and minimal movement during breastfeeding, while controls showed the quiet-alert state ideal for breastfeeding.

Further, the oral reflex assessment and oro-motor examination revealed weak reflexes in 10 neonates (7 in the VHRG and 3 in the HRG) and a mild deviation in the lips in four neonates

of the VHRG. All these four neonates also demonstrated weak oral reflexes. The medical history of these neonates revealed that all of them were preterm associated with low birth weight. The weak reflexes in the VHRG could be attributed to the lack of maturation in the synchronized activities involved in the sucking and swallowing mechanism in these preterm neonates (Allen & Lipkin, 2005; Slattery, Morgan & Douglas, 2012; Lau, 2015). According to Bingham, Ashikaga, and Abbasi, (2010), premature newborns who are ventilated can exhibit significantly poor sucking ability. Persistent, vigorous, weak, or unsymmetrical responses are closely-linked with neurological impairment in full term (Capute, Accardo, Vining, Rubenstein, Walcher, Harryman, & Ross, 1978) and high-risk newborns (Zafeiriou, 2004). Sohn, Ahn, and Le (2015) assessed primitive reflexes in 63 Korean high-risk newborns. They found that 36% of them presented with an abnormal or absent sucking reflex, which was attributed to their clinical condition, such as difficult respiration and decreased mental status.

In addition, it was found that drooling was present in 4.4% of the participants in HRG and 11.1% of the participants in VHRG. This could indicate weakness in the oral structures. None of the participants in the TG had drooling.

## **II Feeding history**

A detailed feeding history was obtained from the mothers of all participants. The questions were related to maternal perception of feeding and/or swallowing problems, feeding method, use of any special feeding aids, feeding duration and frequency, eye contact during feeding, maternal perception of feeding cues and maternal satisfaction with the feeding process.

In an attempt to obtain an understanding about maternal perception of feeding and swallowing difficulties, the answer to the question, “do you feel your child has feeding and



swallowing difficulties?” was elicited from the mothers. 24.2% and 9% of the mothers in the VHRG and HRG respectively reported that their neonates had feeding/swallowing difficulty. This could be due to NICU stay of the VHRG and HRG participants associated with tube feeding due to multifactorial high-risk factors and lesser exposure to the breast feeding. However, 5.3% of the mothers of TG participants also reported that their new-borns had feeding and swallowing difficulties. This could be because of anxiety of the mothers towards feeding their children in the initial feeding sessions (Arts-Rodas & Benoit, 1998). Additionally, in literature, feeding and swallowing related issues in typical children were also reported (Field, Garland, & Williams, 2003). However, serious feeding and swallowing issues are rare in healthy typically developing children (Borowitz & Borowitz, 2018).

With respect to the feeding method, it was found that 80% of the neonates in the TG were breastfed. However, 20% of them were fed using other means such as with a spoon/paladai/medicine dropper/gauze, in addition to breast feeding. Among the HRG, 50.7% of them were breastfed, while 37.3% were fed using other means (spoon/paladai/medicine dropper/gauze), in addition to breast feeding. A small percentage of neonates in the HRG who could not be breastfed, were on paladai/bondla feeding (3%), spoon feeding (1.5%), intravenous feeding (total parenteral nutrition) (1.5%) and nasogastric tube feeds (enteral nutrition) (1.5%). Whereas in the VHRG, only 19.4% of the neonates were exclusively breastfed and 27.8% of them were fed using a spoon/paladai/medicine dropper/gauze, in addition to breast feeding. Compared to the HRG, a greater percentage of the VHRG could not be breastfed and were on other methods of feeding such as paladai feeding (19.4%), spoon feeding (5.5%), nasogastric tube feeding (enteral nutrition) (8.4%) and intravenous method (total parenteral nutrition) (8.4%).

Table 4.1 depicts the different methods of feeding and the percentage of neonates fed through these methods in the three groups of participants.

Table 4.1

*Percentage of different methods of feeding used across the three groups of participants*

<b>Method of feeding</b>	<b>Percentage of participants</b>			
	<b>(N = 178)</b>	<b>TG (N=75)</b>	<b>HRG(N=67)</b>	<b>VHRG(N=36)</b>
Breast feeding		80	50.7	19.4
Breast & spoon / paladai / dropper / gauze feeding		20	37.3	27.8
Paladai feeding		0	3	19.4
Spoon feeding		0	1.5	5.5
IV & Breast feeding		0	3	2.7
IV & Paladai feeding		0	1.5	8.4
NG tube feeding		0	1.5	8.4
IV		0	1.5	8.4

Note\*: TG-Typical group; HRG-High risk group; VHRG-Very high risk group

The finding that twenty percentage of the neonates in the TG had to be supplemented with other types of feeding could attributed to the insufficient lactation as reported by the mothers, sore/burning nipples and deformities in the size and shape of nipples such as flat and

inverted nipples. The problems with the nipples could have made the latching difficult. Gianni, Bettinelli... & Morniroli (2019) studied 552 investigated breastfeeding difficulties experienced by mothers in the first months after delivery. They found that around 70.3% of mothers experienced breastfeeding difficulties, due to cracked nipples, perception of insufficient amount of milk, pain, and fatigue, which occurred within the first month. However, since all the neonates in the TG were on breast feeds, this indicated that they did not have any difficulty in sucking. Borowitz and Borowitz (2018) also reported that serious feeding and swallowing issues are rare in healthy typically developing children.

Further in the HRG and VHRG, a greater percentage of neonates were also fed through other methods. The use of these supplementary feeding methods in these neonates indicates their difficulty in suckling (weak suck) at the breast and/or swallowing due to the presence of associated medical problems. This finding is in concordance with the results reported by Flint, New and Davies (2016), who also reported a poor sucking, latching and suck-swallow-breath coordination issues in children with high risk factors. Jadcherla (2016) also reported that dysphagia is common among high risk neonates with risk factors such as premature birth, low birth weight, birth asphyxia, congenital anomalies and other neurological damage categories. The commonly reported neonatal dysphagic symptoms were latching related difficulties, delayed sucking, lack of rhythmic sucking and tongue movements, reduced extraction of bolus, regurgitation, delayed initiation of pharyngeal phase of swallow, silent aspiration, penetration, failure of peristaltic movements, arching, gagging, irritability, pooling, wet gurgly voice/breathing, coughs while feeding, stridor, discoordination in the pharyngo-upper esophageal sphinctric movements, apnea, feeding related bradycardia, desaturations and other cardiorespiratory events. Jadcherla et al. (2009) also concluded that the dysfunctional neuromotor mechanisms may be responsible for

neonatal dysphagia or its consequences. Slattery et al. (2012) found early sucking and swallowing problems in consistent proportion of infants (35-48%) with varied aetiologies of neonatal brain injury.

Mercado-Deane et al. (2001) also found that the incidence of swallowing dysfunction was significant in premature infants and in those with bronchopulmonary dysplasia, congenital heart disease, esophageal atresia or tracheoesophageal fistula, various syndromes and neurological abnormality. A pilot study by Ferrara, Kamity et al. (2018) also reported pharyngeal phase dysphagia in the preterm infants. Krüger et al. (2019) found that 89.2% of the infants with hypoxic-ischemic encephalopathy had at least one symptom of oropharyngeal dysphagia. 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. Han, Shin and Jeon (2020) also concluded that preterm infants with gestational weeks or with longer ventilation duration are at a higher risk of aspiration.

It was also found that 4.5% and 6.1% of mothers in the HRG and VHRG used special feeding aids to feed their new-borns. They used a special feeding aid called “nipple shield” which could be attached to the breast for feeding. The nipple shield can help the neonates to create suction and position the nipple in a way they may not yet be strong enough to do themselves. The shield holds the nipple in an extended position, ideal for breastfeeding, and allows the new-born to pause and breathe without having to reposition afterwards. This additional use of nipple shield over the natural nipple of the mothers may be due to variations in the nipple type in terms of size and shape such as flat and inverted nipple. It has been reported that the use of nipple shields are common among mothers with flat nipples and is used in the babies who fail to latch effectively on to the breast, especially in the first few days after birth

(Chow, Chow...& Popovic, 2016). According to Meier, Brown, Hurst, Spatz, Engstrom, Borucki, and Krouse (2000), the immature feeding and swallowing behaviours such as weak and ineffective sucking can be compensated by the nipple shield.

The information regarding the feeding duration and frequency was also obtained from the three groups of participants. Table 4.2 depicts the feeding duration and frequency across the three groups. The data revealed that most of the neonates among the TG (40%) and HRG (46.2%) had a feeding duration of 10 to 20 minutes. A small percentage of infants in both these groups (1.3% and 4.5% respectively) also had a feeding duration as long as 30 minutes. However, 66.7% of the VHRG had a feeding duration of less than 10 minutes. None of the participants in the VHRG had feeding duration beyond 20 minutes.

These findings in TG and HRG can be supported by the findings of Mohrbacher (2010), who reported that the average feeding duration in the neonatal period is 20 to 45 minutes. A few of them exhibited feeding duration which was shorter and/or longer than 20 minutes. This could be attributed to the variations in sucking strength, which affects how rapidly they empty a breast (e.g., Pollitt, Gilmore, & Valcarcel, 1978).

A large majority of the neonates in the VHRG exhibited a shorter feeding duration, which could be attributed to the lethargy and fatigue associated medical problems. This group had significantly greater feeding issues as revealed through the NDST tool administered as a part of this study (details provided in the subsequent sections).

Neonatal dysphagia is commonly reported in high risk neonates with risk factors such as premature birth, low birth weight, birth asphyxia, congenital anomalies and other neurological damage categories (Zehetgruber et al., 2014; Jadcherla, 2016). Minde, Perrotta, and Marton in

1985 also found that neonates with very low birth weight were disorganized, with a weak suck, poor lip seal, and problems with the co-ordination of sucking, breathing and swallowing.

It has also been reported that neonates with lower birth weight and who are born prematurely may lack the strength to suck effectively (Institute of Medicine, National Academy of Sciences 1991), which can lead to a longer feeding duration. Geddes, Chooi, Nancarrow, Hepworth, Gardner and Simmer (2017) reported that weaker strength of sucking in preterm and low birth weight new-borns than the full-term babies. However, this finding was not seen in the VHRG in the present study as the feeding duration was shorter than 20 minutes. Jadcherla (2016) also reported that some neonates could have shorter feeding duration (less than 5 minutes) or longer (more than 30 minutes). This variation could be attributed to the under medical condition which could impact the swallowing physiology in different ways.

The data with respect to feeding frequency indicated that majority of the mothers fed their neonates in all the three groups every two hours on an average. This may be due to the counselling and common feeding tips provided by the health care professionals, who insist on feeding young new-borns every two hours. Cobb and Chiu (2012) also reported that new-borns of adequate gestational age were breast fed for every two to three hours with feeding duration was about 10 to 20 minutes on average. A study by Kent, Prime, and Garbin (2010) also revealed that the average feeding frequency for new-borns in 24 hours was 8 to 12 times (or more times), which is in concordance with the present study.

However, there were neonates in all the three groups who were fed every one hour or every thirty minutes. This could be attributed to several reasons. Breasts can vary in the capacity of storing milk. Infants of women with low storage capacity may need to feed their infants more

often to remove the milk and ensure adequate daily intake and production (Daly, Kent, Owens, & Hartmann, 1996). New-borns can also vary greatly in the amount of milk (or formula) they consume during a single feed (e.g., Pollitt et al., 1978). As a result, some babies require more frequent feedings to achieve the same daily caloric intake. Other reported conditions which affected breast feeding were breast engorgement, latch pain and mastitis (inflammation in the breast tissue) (Mangesi & Zakarija-Grkovic, 2016).

Table 4.2

*Feeding duration and frequency across the groups*

<b>Components</b>	<b>Percentage of participants</b>		
	<b>TG (75)</b>	<b>HRG (67)</b>	<b>VHRG (36)</b>
<b>Feeding duration</b>			
<b>&lt;10min</b>	34.7	26.9	66.7
<b>10-20 min</b>	40	46.2	24.2
<b>21-30 min</b>	25.3	26.9	0
<b>NA</b>	0	0	9.1
<b>Feeding Frequency</b>			
<b>0.5 hour</b>	10.7	6	12.1
<b>1 hour</b>	32	23.9	6.1
<b>2 hours</b>	57.3	70.1	72.7
<b>NA*</b>	0	0	9.1

Note\*: TG-Typical group; HRG-High risk group; VHRG-Very high risk group

NA- Not applicable as the children were on IV feeding

Generally, eye gaze develops within seven hours of birth and this developmental milestone has significance in the association of food and feeder (Meltzof & Brooks, 2007). Most

mothers indicated that their neonates were able to make eye contact. However, the comparison across the groups revealed that greater number of neonates (24.2%) in the VHRG did not make eye contact during feeding compared to TG (8%) and HRG (6%). On reviewing the medical history of these neonates, it was found that there were no specific risk factors associated to poor eye contact. Krüger, Kritzinger, and Pottas (2019) also found that 89.2% of the infants with hypoxic-ischemic encephalopathy with oropharyngeal dysphagia had significantly more closed eyes and minimal movement during breastfeeding.

The information pertaining to the perception of hunger and satiety cues exhibited by the child for the initiation and termination of feeding respectively were elicited from the mothers. The results indicated that the majority of the mothers (96.6%) in all the groups were able to perceive the hunger and satiety cues signalled by their neonates. Only 3.4% of mothers were not able to perceive the feeding cues. This finding is consistent with the studies by Gross et al. (2010), and Crow (1977) who stated that most mothers could perceive the hunger and satiety cues exhibited by their infants and acted accordingly in initiating and terminating the feeding sessions (responsive feeding).

It was seen that in all the three groups, crying was the most frequent response with regard to the hunger cues. With respect to the satiety cues, in the typical group, the decreasing/stopping sucking was most frequent among the responses (30.7%), whereas in the high and very high-risk group, pushing nipple from mouth and falling asleep were most frequent response respectively (43% & 47.2%).

These findings are in concordance with the study by Gross et al. (2010) who reported crying as a commonly perceived hunger cue by more than 70% of the mothers in typical infants.



Further, the findings with respect to satiety cues are also in concordance with that reported by Crow (1977) and Morris, Roger, and Taper (1983). Hodges, Hughes, Hopkinson and Fisher (2008) reported “crying/fussy, increased sucking/mouthing, rooting, on demand” and “nipple detachment / release / spits, stops and refuses” as frequent feeding initiation and termination cues respectively for typical children less than 3 months. In the present study, though crying was a frequently seen hunger cue, sucking and rooting were perceived by lesser number of mothers. The finding of nipple detachment/stopping sucking was also in agreement with the study by Hodges et al. (2008).

Falling asleep was the most frequent response for the satiety cue in the high-risk group. This could be attributed to the underlying medical conditions which could have lead to lethargy and fatigue in these children, making them to fall asleep. The studies on maternal perception and interpretation of feeding cues (hunger and satiety) in high risk infants are scarce and hence a direct comparison with the present study cannot be drawn.

As a part of assessing the maternal and infant satisfaction in the feeding process, the information relating to questions such as “Are you satisfied with the quantity of milk intake?” (MSQ1), “Are you happy with the baby’s current weight?” (MSQ2) and “Does your child seem satisfied / calm after feeding?” (MSQ3) were elicited. It was seen that 81.4% of the mothers in the TG and 83.5% of the mothers in the HRG were satisfied with the quantity of milk intake. However, in the VHRG, only 63.8% of the mothers were satisfied. Similarly, 94.6% and 97% of the mothers in the TG and HRG respectively were happy with their children’s current weight gain, whereas, only 72.4% of the mothers in the VHRG were satisfied. Further, 86.6% and 79.1% mothers in the TG and HRG respectively reported that their children were satisfied and calm after feeding, whereas in the VHRG, less than half of the mothers (44.4%) expressed that

their children were satisfied after feeding. A small percentage of participants in the HRG and VHRG also reported that they could not judge these aspects.

It was evident that the mothers of TG and HRG were more satisfied with the feeding process than VHRG. These differences may be attributed to the presence of multiple medical conditions and variations in the feeding methods used in the VHRG. In VHRG, use of enteral and parenteral feeding methods, in particular would have contributed to the dissatisfaction among the mothers of neonates in the VHRG compared to the mothers of neonates in the TG and HRG.

### **III Findings on feeding and swallowing assessment**

The fifth section of the NDST was administered on 178 mothers of neonates, which consisted of 17 questions (Q1 to Q17) with binary choice (yes/no) scoring system. Higher scores on this indicated greater feeding and/or swallowing difficulties. The NDST was re-administered on all the neonates at the end of one month to assess if there were any changes in the feeding and swallowing functions over time. NDST was again re-administered at the end of four months on those neonates in the HRG and VHRG. In addition, the NDST was administered on a different group of participants in the age range of 0.6 to 4 years with (N=30) and without dysphagia (N=30) for establishing its sensitivity and specificity.

The overall score for each participant was calculated by adding the individual score on each question/item based on the rating scale. The obtained data was tabulated and subjected to appropriate statistical analysis. The following statistical procedures were carried out using SPSS software (version 20.0).

- Descriptive statistics to obtain mean, median and standard deviation across the groups.
- Cohen's kappa coefficient to determine the inter-rater reliability.
- Mann Whitney U test to check any significant differences between the clinical and the control groups.
- Chi-square test to measure the level of significant association between mother's response on each test item of NDST and the groups.
- Kruskal-Wallis test to detect the presence of significant differences if any, across all the three groups (TG, HRG & VHRG).
- Wilcoxon test to identify significant differences if any, between mother's responses on first (within 5 days of birth) and second administration (at the end of one month) of NDST across the groups.

The results obtained using all the above statistical procedures have been presented and discussed under the following sections:

Section I: Clinical validity of NDST

Section II: Sensitivity and Specificity of NDST

Section III: Inter-rater reliability of NDST

Section IV: Critical test items of NDST

Section V: Establishment of scoring and interpretation algorithm for NDST

## **Section I: Clinical validity of NDST**

The finalized version of the NDST was administered on participants of the TG, HRG and VHRG. The information obtained using the section E of NDST on feeding and swallowing screening was tabulated and analyzed using SPSS (version 20.0) and the results have been presented and discussed below.

### **a) Comparison across groups on the first and second administration of NDST**

To identify the presence of feeding and swallowing difficulties, the section E was administered within the first five days of their birth and at the end of one month. The median scores were used to compare the groups because the standard deviation was greater than the mean scores. The mean, median and standard deviation of the three groups for the first administration of the NDST have been depicted in the table 4.3. In the first administration, the median scores of VHRG (M=3.00) was higher than the HRG (M=1.00) and TG (M=0.00). The greater median scores of the VHRG indicated that they had greater feeding and swallowing difficulties, in comparison to the other two groups. Kruskal-Wallis test revealed a very high significant difference at  $p < 0.001$  level across all three groups ( $\chi^2=30.21$ ,  $df=2$ ,  $p=0.00$ ). To check between which groups a significant difference existed, Mann-Whitney test was performed. The results revealed that there was a highly significant difference ( $p < 0.01$ ) between TG & HRG; TG & VHRG and between HRG & VHRG. The  $Z$  values have been depicted in table 4.4.

A similar pattern of results was obtained on comparing the median of overall scores obtained on the second assessment using NDST on the same participants at the end of a month. That is, the median score of VHRG (M =3.00) was higher than HRG (M =0.00) and TG (M =0.00). The mean, median and standard deviation of the three groups for the second administration of the NDST have been depicted in the table 4.3. Kruskal-Wallis test revealed a

very high significant difference ( $p < 0.001$ ) in the second administration of NDST across all three groups ( $\chi^2=75.87$ ,  $df=2$ ,  $p=0.00$ ). Mann-Whitney test revealed a highly significant difference ( $p < 0.01$ ) between TG & HRG; TG & VHRG and also between HRG and VHRG. The /z/ values have been depicted in table 4.4.

Table 4.3

*Mean, median and standard deviation (SD) of three groups on the first and second assessment using NDST*

Assessment	TG		HRG		VHRG	
	Mean±SD	Median	Mean±SD	Median	Mean±SD	Median
First Assessment	0.79±1.48	0.00	1.67±1.97	1.00	3.34±2.56	3.00
Second assessment	0.05±0.28	0.00	0.37±0.88	0.00	3.09±2.58	3.00

Note-TG-Typical group; HRG-High risk group; VHRG-Very high risk group

Table 4.4

*Results of Mann-Whitney test across groups on the first and second administration of NDST.*

Assessment	z  value		
	TG & HRG	TG & VHRG	HRG & VHRG
First assessment	3.36**	5.20**	3.06**
Second assessment	2.93**	7.96**	6.15**

Note: at \*\*  $p < 0.01$ ; TG-Typical group; HRG-High risk group; VHRG-Very high risk group

From the above findings, it is evident that the feeding/swallowing difficulties were significantly greater for the VHRG followed by HRG and TG. A similar trend of higher feeding and swallowing difficulties in very high-risk neonates was reported by Viviers et al. (2016) and Lee and Seo (2016). The higher percentage of difficulty in VHRG than HRG could probably be attributed to the type of risk factors associated with it. That is, VHRG contained neonates with risk factors that are commonly associated with neonatal dysphagia such as extremely low birth weight (ELBW), prematurity, congenital anomalies, Respiratory Distress Syndrome (RDS), Hypoxic Ischemic Encephalopathy (HIE) and convulsions. Some of the infants also exhibited more than one risk factor as well. Lee, et al. (2011), Ferrara, et al. (2018), Krüger et al. (2019) also reported dysphagia in low birth weight, preterm and infant with HIE respectively. Slattery, Morgan, and Douglas in 2012 found early sucking and swallowing problems in consistent proportion of infants (35-48%) with varied aetiologies of neonatal brain injury.

This finding is also in agreement with the results reported by the Jadcherla (2016) and Farneti and Genovese (2017). Jadcherla (2016) also reported a list of associated feeding and swallowing difficulties in high risk neonates such as latching problems, delayed initiation of sucking, gagging, choking and respiratory disturbances. However, in the present study, additional difficulties such as shorter span of feeding, forward loss of fluid from the oral cavity (oral pooling), stimulation initiated sucking and frequent vomiting were also reported. Further, the study by Farneti and Genovese (2017) reported additional feeding and swallowing difficulties in high risk new-borns such as weak feeding, incoordination between the rhythm of sucking and swallowing, changes in the pattern of breathing during feeding, dehydration, marked irritability and lethargy during feeding.

### b) Comparison across first and second assessment for all the groups

The median scores of first and second administration of NDST were compared for all the three groups. It was observed that the scores had reduced considerably in the participants of the TG and HRG, which indicated that their feeding and swallowing difficulties had reduced with time. However, there was no reduction in the median scores in the VHRG, which indicated that their feeding and swallowing difficulties persisted. The median NDST scores of all the three groups on the first and second administration of NDST have been depicted graphically in the figure 4.1.

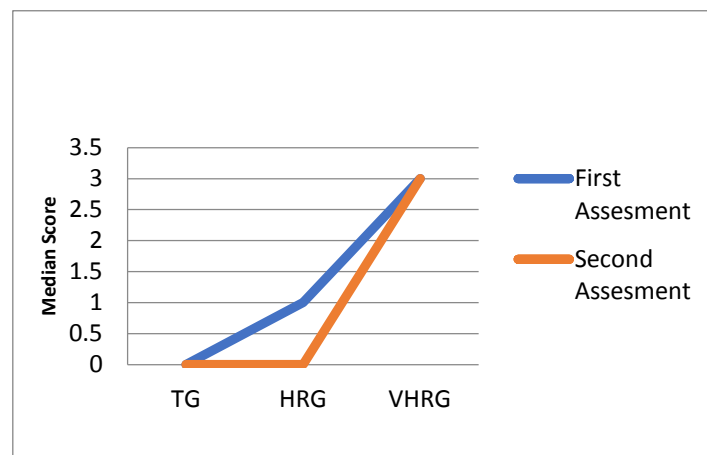


Figure 4.1: Median score of the three groups on the first and second assessment using NDST.

Using the Wilcoxon test, all the three groups were compared separately across the first and second NDST assessment. The Wilcoxon test results revealed a very high significant difference ( $p < 0.001$ ) between both the assessments for TG ( $|z|$  value = 4.27,  $p=0.00$ ) and HRG ( $|z|$  value = 5.45  $p=0.00$ ). However, for VHRG, no significant difference was observed between the first and second assessment ( $|z|$  value = 1.18  $p=0.24$ ). This lack of significant difference in

the VHRG clearly indicated the persistence of difficulties in feeding and swallowing even at the end of the first month.

The finding that VHRG continued to have difficulty in feeding and swallowing functions compared to the other groups could be attributed to the more severe risk factors in the VHRG such as respiratory distress syndrome, hypoxic ischemic encephalopathy, convulsions, congenital anomalies, prematurity and extremely low birth weight, which impedes the development of feeding and swallowing mechanism. The participants in the other groups such as HRG and TG had overcome their difficulties in feeding and swallowing functions to a great extent. This improvement in the feeding and swallowing function in TG and HRG could be due to the presence of less severe risk factors.

**c) Distribution of mother's responses on each test item in the first assessment using NDST across the groups**

The responses obtained from the mothers on the first administration of NDST across the groups have been depicted in table 4.5. It was seen that greater than 85% of the neonates in the TG did not experience difficulties with feeding for any of the questions on the NDST. However, 13.3% of neonates had frequent hiccups after feeding (Q16) and 12% vomited after feeding (Q17). A very negligible percentage of neonates had difficulties with other questions on the NDST such as difficulty in latching and staying fixed to the breast.

Whereas in the HRG, 26.9% of neonates required some stimulation to initiate a suck (Q6), 22.4% experienced frequent hiccups after feeding (Q16), 17.9% took longer time to suck (Q5), 16.4% had difficulty in staying fixed to the breast and vomited after feeding (Q 4 and 17), 13.4% had difficulty in latching (Q3), and 11.9% experienced



coughing/choking/gagging/tearing/turning blue (Q14). All other difficulties were present in less than 10% of the participants.

However in the VHRG, 42.4% of the neonates had frequent coughing/choking/gagging/tearing/turning into blue (Q14), 30.3% had short period of feeding (less than 5 minutes) and fluid leak from the mouth (Q10 & Q12); 27.3% had difficulty in staying fixed to the breast (Q4); 21.2% had delayed sucking after latching and breathing difficulties/loud breathing noises during or after feeding (Q5 & Q9), 18.2% had difficulty in latching (Q3), required stimulation to initiate suck (Q6), delayed initiation of swallow (Q7) and vomiting after feeding (Q17), all of which were reported only by a negligible percentage of mothers in the other two groups.

It was seen that less than 10% of the neonates among all the three groups had difficulty in turning head to the mother for feeding (Q1), difficulty in opening the mouth in response to the nipple (Q2), refusal to drink milk (Q8), prolonged feeding duration (Q11), nasal regurgitation (Q13), and wet/gurgly voice after feeding (Q15).

A small percentage of participants in the HRG and VHRG reported that they could not judge the feeding difficulty in their new-borns as they were unsure, therefore they did not answer within the binary choice. That is, 6% (in HRG) and 45.5% (in VHRG) of the mothers could not answer the questions Q1 to Q6 as their neonates were on alternate mode of feeding (questions were more appropriate only for breast feeding). 1.5 % of the participants in HRG and 9.1% of the participants in VHRG could not answer the questions Q7 to Q17, since it focused on aspects of feeding and swallowing during or immediately after feeding. This could be attributed to the lesser time of mother-child interaction consequent to the NICU stay, alternative modes of

feeding such as tube feeding with lesser or no exposure to breast feeding, consequent to the presence of serious medical conditions.

The results of the chi-square test revealed an association between the responses obtained and the groups for all the questions on NDST, which indicated that all the questions were significant in the classification of the groups. There was a very high significant association ( $p < 0.001$ ) for Q1, Q2, Q3, Q4, Q5, Q6, Q7, Q9, Q10, Q12 and Q14 and a significant association ( $p < 0.05$ ) for Q8, Q11, Q13, Q15, Q16 and Q17 of NDST, between the responses obtained and the groups. The chi-square value have been depicted in the table 4.5.

Table 4.5

*Distribution of mother's responses obtained on the first assessment using NDST in percentage and the results of chi-square test.*

Q.	TG		HRG			VHRG			$\chi^2(df=4)$
	Yes %	No %	Yes %	No %	Nil %	Yes %	No %	Nil %	
Q1	1.3	98.7	1.5	92.5	6	3	51.5	45.5	52.57***
Q2	0	100	7.5	86.5	6	9.1	45.4	45.5	60.24***
Q3	9.3	90.7	13.4	80.6	6	18.2	36.3	45.5	56.55***
Q4	9.3	90.7	16.4	77.6	6	27.3	27.2	45.5	63.57***
Q5	6.7	93.3	17.9	76.1	6	21.2	33.3	45.5	61.872***

Q6	8	92	26.9	67.1	6	18.2	36.4	45.5	63.58***
Q7	1.3	98.7	3	95.5	1.5	18.2	72.7	9.1	23.99***
Q8	1.3	98.7	3	95.5	1.5	0	90.9	9.1	9.99*
Q9	0	100	1.5	97	1.5	21.2	69.7	9.1	35.83***
Q10	6.7	93.3	7.5	91.0	1.5	30.3	60.6	9.1	24.41***
Q11	1.3	98.7	3	95.5	1.5	0	90.9	9.1	9.99*
Q12	2.7	97.3	10.4	88.1	1.5	30.3	60.6	9.1	28.30***
Q13	1.3	98.7	1.5	97	1.5	6.1	84.8	9.1	11.61*
Q14	2.7	97.3	11.9	86.6	1.5	42.4	48.5	9.1	41.84***
Q15	1.3	98.7	0	98.5	1.5	3.0	87.9	9.1	10.72*
Q16	13.3	86.7	22.4	76.1	1.5	1.2	69.7	9.1	11.34*
Q17	12	88	16.4	82.1	1.5	18.2	72.7	9.1	10.01*

Note-\*\*\* p < 0.001, \* p < 0.05; TG-Typical group; HRG-High risk group; VHRG-Very high-risk group

The findings indicated that the neonates in the VHRG demonstrated feeding and swallowing difficulties on 10 questions on the NDST. It was seen that maximum percentage of them had coughing / gagging / choking / tearing or watery eyes/ turning to blue during or immediately after feeding, anterior spillage of fluid and shorter feeding span, which were greater than that seen in the HRG and TG. The other most frequently observed feeding and swallowing

difficulties in the VHRG included difficulty in staying fixed to the breast, taking longer time to suck, breathing difficulties during and after feeding, which were again greater than that seen in HRG and TG. These findings indicated that they had difficulties with the pharyngeal and the oral phase of swallow. It is interesting to note that the response to the Q15 on wetness/gurgliness in voice after feeding was negligible, though this was related to choking during feeding. This could be because of that fact that the mothers were not yet sensitive to the differences in voice of their new-borns.

The HRG, on the other hand, experienced difficulties on a lesser number of questions (8) on the NDST. They also differed from the other groups on the nature of difficulties. Though a large majority of VHRG had coughing/choking and shorter span of feeding, lesser percentage of HRG had these difficulties. Instead they had difficulties with sucking, i.e., they had difficulty in latching, staying fixed to the breast, needed longer time to suck and required stimulation to initiate it. They also had frequent hiccups and vomited after feeding. This indicated that they had more difficulties in the oral phase, rather than the pharyngeal phase of swallow.

Weak sucking due to weakness of oral muscles, in-coordination of sucking, swallowing and breathing leading to choking, apnea, increased respiratory rate, loud breathing noises and aspiration has been reported in high risk neonates (Laitman & Reidenberg, 1993). Difficulty in initiating sucking and swallowing, delayed sucking, latching related difficulties, labial spillage or anterior spillage of liquid, shorter (less than 5 minutes), silent aspiration, penetration, gagging, irritability, pooling, wet gurgly voice/breathing, coughs while feeding, stridor, and frequent vomiting have also been reported by Jadcherla (2016).

The presence of cough/choking/ tearing/turning blue in the VHRG group (greater extent) and HRG (lesser extent) indicated aspiration, which occurs due to in-coordination among breathing, sucking and swallowing sequence. Aspiration has been reported to range from 25% to 73% for infants with swallowing dysfunction (Mercado-Deane et al., 2001). Newman et al. (2001) reported that ~85% of children exhibited aspiration. Mercado-Deane et al. (2001) assessed the incidence of swallowing dysfunction in infants with vomiting or respiratory symptoms. They found that 13.4% had swallowing dysfunction which included aspiration and penetration.

Jadcherla et al. (2009) found that 30% had nasopharyngeal reflux, 35% experienced pooling, 35% had delayed swallow, 55% had aspiration, and 90% experienced laryngeal penetration using videofluoroscopy. However, in the current study nasal regurgitation was seen only in 6% of the VHRG and 1.5% of the HRG. This could be because of the differences in the underlying aetiology in both the studies.

In the TG, the feeding difficulties were seen only on two questions on NDST, which were frequent hiccups and vomiting after feeding. A very negligible percentage of them had difficulty in latching and staying fixed to the breast. Coughing/choking was seen in a very negligible percentage of TG. Gleeson et al. (2011) also reported that aspiration can occur normal infants, however, the same is cleared by the body's defence mechanism. Delzell, Kraus, Gaisie, and Lerner (1999) reported that laryngeal penetration was seen in most normal infants, which could be attributed to the immaturity of the swallowing mechanism, however, this does not lead to aspiration. Borowitz and Borowitz (2018) reported that serious feeding and swallowing issues are rare in healthy typically developing children.

Hiccups were seen to a greater extent in HRG and TG, which could have been caused by overfeeding and also by air trapping in the stomach while swallowing, which has been reported by Ceriani, Fogliani, and Kastermann(2010). However, frequent hiccups are associated with gastro-esophageal reflux disease (GERD) and incomplete synchronization of breathing and swallowing abilities as reported by Whitehead, Jones, Laudiano-Dray, Meek, and Kastermann, (2019). In both these groups vomiting after feeding was also seen, which could have indicated the presence of GERD. Hiccups have also been reported in preterm new-borns due to the immaturity of the gastro-intestinal functions (Morris & Klein, 2000).

**d) Distribution of mother's response on each test item in the second assessment using NDST across the groups**

The second administration of NDST was performed for all the participants of TG, HRG and VHRG at the end of the first month. Mothers' responses to all the 17 questions of NDST were documented and have been depicted in the table 4.6. Results showed that the mothers of the participants in the TG did not report any feeding difficulties. A negligible percentage of participants in this group had issues with Q16 (4%, hiccups) and 14 (1.3%, choking). However, in the HRG, 10.4% neonates had difficulty with Q14 & 16. Only a negligible percentage of them had difficulties on Q 4,9,10,12,13,14,16 &17.

In VHRG, 60.6% of the neonates had frequent coughing/choking/gagging/tearing or watery eyes/turning into blue during or immediately after feeding (Q14); 48.5% had frequent hiccups after feeding (Q16); 45.5% reported leak of fluid from their babies mouth (Q12), 27.3% and 24.3% reported that their babies had difficulty in staying fixed to the breast and delayed sucking after latching (Q4 & Q5) and 21.1% had difficulty in initiating a swallow and vomited

after feeding. All the other difficulties were reported by less than 10% of the neonates in the VHRG. However, three participants from VHRG reported the expiry of their new-borns due to associated very high-risk factors and hence were excluded. Further, none of the neonates in all the groups had a prolonged period of feeding and wet/gurgly voice after feeding (Q11 & Q15).

The results of the chi square test revealed a very high significant association ( $p < 0.001$ ) for Q3, Q4, Q5, Q6, Q7, Q12, Q14 and Q16, a high significant association ( $p < 0.01$ ) for Q17 and a significant association ( $P < 0.05$ ) for Q9 and Q13 between the responses obtained and the groups. No association was seen for Q1, Q2, Q8 and Q10. This could be consequent to the decrease in feeding difficulties seen in all the groups. For Q11 and 15, since the responses were 0, association could not be obtained. The chi-square values have been depicted in table 4.6.

Table 4.6

*Distribution of mother's responses obtained on the second assessment using NDST and the results of chi-square test.*

Q.	TG		HRG		VHRG			$\chi^2$	Df
	Yes (%)	No (%)	Yes (%)	No (%)	Yes (%)	No (%)	Nil (%)		
Q1	0	100	0	100	3	94	3	8.71	4
Q2	0	100	0	100	3	94	3	8.71	4
Q3	0	100	0	100	9.1	87.9	3	17.66***	4
Q4	0	100	1.5	98.5	27.3	69.7	3	40.03***	4

Q5	0	100	0	100	24.3	72.7	3	40.83***	4
Q6	0	100	0	100	9.1	87.9	3	17.62***	4
Q7	0	100	0	100	12.1	87.9	0	17.62***	2
Q8	0	100	0	100	3	97	0	4.33	2
Q9	0	100	1.5	98.5	9.1	90.9	0	8.79*	2
Q10	0	100	1.5	98.5	6.1	93.9	0	5.03	2
Q11	0	100	0	100	0	100	0	-	-
Q12	0	100	6	94	45.5	54.5	0	51.60***	2
Q13	0	100	1	99	9.1	90.9	0	6.83*	2
Q14	1.3	98.7	10.4	89.6	60.6	39.4	0	62.39***	2
Q15	0	100	0	100	0	100	0	-	-
Q16	4	96	10.4	89.6	48.5	51.5	0	37.53***	2
Q17	0	100	3	97	12.1	87.9	0	10.23**	2

Note- -\*\*\* p < 0.001, \*\* p < 0.01, \* p < 0.05; TG-Typical group; HRG-High risk group; VHRG-Very high risk group

These findings indicated that the feeding and /or swallowing difficulties in the VHRG reduced with time. That is, neonates had difficulties on seven questions on the NDST, as opposed to difficulties seen across 10 questions during the first assessment. However, it was seen that the percentage of neonates with coughing/choking/tearing during/after feeding increased during the second assessment compared to the first assessment. Also, greater number of neonates



had anterior spillage compared to the first assessment. This could have occurred because of the persistent difficulties in the oral and pharyngeal phase. The frequency of hiccups also increased in the second assessment, possibly due to the difficulty in coordinating between breathing and swallowing. The difficulties such as staying fixed on the breast and taking long time to suck persisted in these children. The delayed initiation of swallow and vomiting after feeding decreased as lesser percentage of neonates demonstrated these problems compared to the first assessment. This could be attributed to the maturational effects.

In the HRG, the neonates had difficulties only on two questions on the NDST, as opposed to eight questions in the first assessment. The hiccups and coughing/choking/tearing during/after feeding, though were present was seen in lesser percentage of neonates compared to the first assessment. Lesser percentage of the neonates of TG experienced issues, which again were lesser compared to the first assessment. The decrease in feeding and/or swallowing difficulties could be attributed to the overall development and maturation of the neurological, respiratory, and gastrointestinal functions.

#### **e) Comparison across HRG and VHRG on the third assessment using NDST**

The third assessment was conducted only on 57 participants of HRG (40) and VHRG (17) at the end of four months to assess their feeding and swallowing abilities. The mean and standard deviation of HRG were  $0.25 \pm 0.79$  respectively. Similarly, mean, and standard deviation of VHRG were  $1.43 \pm 1.76$  respectively. The median scores were used to compare between the groups because the standard deviation was greater than the mean scores. The median of VHRG (M=0.50) was higher than HRG (M=0). The overall NDST median scores obtained in the third administration on the HRG and VHRG were compared using Mann-Whitney test to check for the

presence of any significant difference between these groups. The results revealed that there was a very high significant difference ( $p < 0.001$ ) between HRG and VHRG ( $|z|=3.99$ ).

These findings indicated that the HRG was significantly different from the VHRG on the third assessment, with the VHRG still persisting with greater feeding and /or swallowing difficulties compared to the HRG. This could be due to the more serious and multiple underlying medical conditions that were present in VHRG. In a review, Jadcherla (2016) stated that prematurity, respiratory distress syndrome, epilepsy, low birth weight, and encephalopathy were the predominant risk factors generally associated with neonatal dysphagia.

**f) Distribution of mother's response on each test item in the third assessment using NDST across HRG and VHRG**

Table 4.7 depicts the distribution of mother's responses obtained on the third assessment using NDST for neonates in the HRG and VHRG. It was seen that the feeding and/or swallowing difficulties in the VHRG was restricted to only four questions, as opposed to difficulties across seven questions seen in the second assessment. Specifically, 33.3% of the neonates in the VHRG had coughing/choking/tearing (Q14), 24.2% had anterior spillage difficulties (Q12), 15.2% had hiccups (Q16), 12.1% had difficulty with staying fixed on the breast (Q4). Less than 10% of the neonates had difficulties with the other questions on the NDST. On six questions (Q1,2,6,9,11, and 15), none of the neonates had any difficulties. On the other hand, only a very negligible percentage of participants in HRG had difficulties and these were found only on a few questions, as opposed to greater difficulties on two questions related to coughing/choking/tearing and hiccups seen in the second assessment. The results of the chi-square test revealed a very high

significant association ( $p < 0.001$ ) between the mother's response and the groups for all the test items of the NDST. The chi-square values have been depicted in the table 4.7.

These findings indicated that the feeding and/or swallowing problems reduced at the end of the fourth month in comparison to the second assessment in both groups. This could be attributed to the neurological maturation with age.

Table 4.7

*Distribution of mothers' responses obtained for HRG and VHRG on the third assessment using NDST and the results of chi-square test.*

Q.Code	HRG		VHRG		$\chi^2$	Df
	Yes (%)	No (%)	Yes (%)	No (%)		
Q1	0	85.1	0	90.9	171.74****	4
Q2	0	85.1	0	90.9	171.74****	4
Q3	0	85.1	6.1	84.8	178.69****	6
Q4	1.5	83.6	12.1	78.8	180.44****	6
Q5	0	85.1	3	87.9	175.18****	6
Q6	0	85.1	0	90.9	171.74****	4
Q7	3	97	9.1	90.9	174.48****	6
Q8	3	97	9.1	90.9	174.48****	6

Q9	0	100	0	100	171.74****	4
Q10	0	100	6.1	93.9	178.69****	6
Q11	0	100	0	100	171.74****	4
Q12	3	97	24.2	75.8	190.27****	6
Q13	1	99	9.1	90.9	174.48****	6
Q14	4.5	95.5	33.3	66.7	202.27****	8
Q15	0	100	0	100	171.74****	4
Q16	1.5	98.5	15.2	84.8	183.91****	6
Q17	1.5	98.5	3	97	172.13****	6

Note -\*\*\* p < 0.001, HRG-High risk group; VHRG-Very high risk group  
P.S: Remaining percentages in high risk and very high risk group were no response/  
NIL due to different methods of feeding like spoon feeding, paladai feeding and so on

Using the “Remember and Care” checklist, the development of the infants in the HRG and VHRG was also assessed in terms of their motor, hearing, speech and language abilities. The percentages of infants with delayed milestones in both the groups have been provided in the table 4.8 below. The data revealed that 9.1%, 27.3% and 12.1% of the infants in VHRG presented issues in the development of hearing, motor, speech and language respectively. While in the HRG, only 4.5% and 1.5% of the infants presented with motor, speech and language related issues respectively. In addition, the results of the chi-square test demonstrated a very high significant association ( $p < 0.001$ ) between the three developmental milestones and the groups. The chi-square values have also been depicted in the table 4.8.

Table 4.8

*Percentage of infants with delayed milestones in both the groups and the results of chi-square test*

Milestone	HRG		VHRG		$\chi^2$ (df=2)
	Normal	Delayed	Normal	Delayed	
Hearing	100	0	90.9	9.1	13.13***
Speech & Language	98.5	1.5	87.9	12.1	12.86***
Motor	95.5	4.5	72.7	27.3	27.65***

Note - \*\*\*  $p < 0.001$ ; HRG-High risk group; VHRG-Very high-risk group

The results of the assessment of development of infants in both the groups revealed that greater number of infants in the VHRG had delayed development compared to the infants in the HRG in hearing, speech and language and motor domains. These delayed developmental milestones might be due to the presence of prenatal and peri-natal high-risk factors in these children. In the literature, pre-conceptional risk factors, maternal infection and chronic illness, maternal nutritional deficiencies, meconium aspiration, birth asphyxia, prematurity and low birth weight were reported as higher risk factors for delays in developmental milestones (Sharma, Masood, Singh, Ahmad, Mishra, Singh, & Bhattacharya, 2019). Therefore, the presence of very high-risk factors in the participants of the VHRG might be the basis for the delay in their developmental milestones.

The motor milestones were delayed in maximum percentage of children in both the groups. The delay in the motor development could have contributed to most of the feeding and or swallowing problems seen in both the groups as feeding and swallowing is a motor activity and involves the movements of several muscles. This finding is in coherence with study by Sullivan, Lambert, Rose, Ford-Adams, Johnson, and Griffiths (2000) who reported that children with severe motor involvement had greater deficits in feeding and swallowing. Malas, Trudeau, Chagnon, and Farland (2015) also reported a higher percentage of feeding and swallowing disorders in children with language and motor impairments. Therefore, feeding-swallowing difficulties may be suggestive of underlying and possibly subtly represented motor impairments with distributed impacts throughout feeding - swallowing and speech - language systems (Noterdaeme, Mildenberger, Minow, & Amorosa, 2002; Nip, Green, & Marx, 2011).

**g) Comparison between two other groups (clinical and typical) on NDST**

The NDST was administered on another clinical group (children with a confirmed diagnosis of oro-pharyngeal dysphagia in the age range of 0.6 to 4 yrs) and an age and gender matched typical group (typically developing children) and the scores were compared across the groups. The information on NDST was elicited retrospectively, i.e. whether the children had any feeding problems in the first month of their life. Descriptive statistics was computed to obtain the mean, median and standard deviation. The median scores were used to compare the groups because the standard deviation was greater than the mean scores. The median of typical and the clinical group were 0 and 4 respectively. The mean and SD values of typical group were 0.23 and 0.50 and clinical group were 4.63 and 2.17 respectively. Further, to check the presence of any significant difference between these groups on NDST scores, Mann-Whitney test was

performed. The results revealed the presence of a very high significant difference between the typical and clinical group ( $|z|=6.88$ ,  $p < 0.001$ ).

These findings indicated that the clinical group had significantly greater feeding difficulties than the typical group. This could be attributed to the delayed development and neurological issues that they exhibited. Twenty-one of them had an overall developmental delay and 9 had cerebral palsy. Feeding difficulties right from birth has been reported in children with cerebral palsy. 57% of the infants with CP were reported to have sucking problems, 38% had swallowing problems within 12 months of life and 80% of the infants were fed nonorally at least once (Reilly, Skuse, & Poblete, 1996). This finding is also in coherence with van den Engel-Hoek, de Groot, de Swart, and Erasmus(2015) where they reported neonatal onset of feeding and swallowing problems in children with risk factors. Oropharyngeal dysphagia has been reported to be present in 90% of children with CP (Benfer, Weir, Bell, Ware, Davies & Boyd, 2012; Benfer, Weir, Bell, Ware, Davies & Boyd, 2013).

#### **h) Distribution of mother's responses on each test item across the clinical and typical group**

The responses of the 60 mothers of participants in both the groups on each item of NDST have been depicted in the table 4.9. The results indicated that the more than 10% of participants in the clinical group had feeding and swallowing difficulties across 14 items on the NDST. Maximum percentage of children (56.7%) had difficulty in staying fixed to the breast and took a long time to suck after latching. Fifty percentage of children had difficulty in latching and required stimulation to initiate a suck, 46.7% experience frequent hiccups after feeding, 36.7% vomited after feeding and had anterior spillage of fluid, 33.3% had

coughing/choking/tearing/turning blue, 23.3% had delayed initiation of swallow, 13.3% had difficulty in opening mouth during feeding, short period of feeding and nasal regurgitation and 10% had gurgly voice and difficulty in turning head towards the breast during feeding. The other problems were present in a negligible percentage of children in the clinical group.

The children in the typical group did not experience any feeding difficulties during their infancy as reported by their mothers on most questions of the tool. Only less than 10% of the participants in the typical group had food refusal (Q8-3.3), short span of feeding (Q10-6.7%), fluid leak from the mouth (Q12-3.3%), frequent hiccups (Q16-6.7%) and vomiting after feeding (Q17-3.3%) during their neonatal period.

Chi-square test results revealed a very high significant association for Q3, Q4, Q5, Q6, Q12, Q14, Q16 and Q17 ( $P \leq 0.001$ ) and a significant association for Q2 and Q13 ( $p > 0.001 \leq 0.01$ ) between the mother's response and groups. However, there was no association seen on the other questions of the NDST. The chi-square values have been depicted in table 4.9.

Table 4.9

*Distribution of mothers' responses obtained for the two groups.*

Q. Code	Typical group (N = 30)		Clinical group(N = 30)		$\chi^2$	Df
	Yes (%)	No (%)	Yes (%)	No (%)		
Q1	0	100	10	90	3.16	1
Q2	0	100	13.3	86.7	4.29*	1
Q3	0	100	50	50	20.00***	1
Q4	0	100	56.7	43.3	23.72***	1



Q5	0	100	56.7	43.3	23.72***	1
Q6	0	100	50	50	20.00***	1
Q7	0	100	23.3	76.7	7.93	1
Q8	3.3	96.7	6.7	93.3	0.35	1
Q9	0	100	3.3	96.7	1.02	1
Q10	6.7	93.3	13.3	86.7	0.74	1
Q11	0	100	3.3	96.7	1.02	1
Q12	3.3	96.7	36.7	63.3	10.42***	1
Q13	0	100	13.3	86.7	4.29*	1
Q14	0	100	33.3	66.7	12.00***	1
Q15	0	100	10	90	3.16	1
Q16	6.7	93.3	46.7	53.3	12.27***	1
Q17	3.3	96.7	36.7	63.3	10.42***	1

Note- -\*\*\*  $p \leq 0.001$ , \*  $p > 0.001 \leq 0.01$

The findings indicated that children in the clinical group had feeding and swallowing difficulties across almost all questions on the NDST than the control group. The type of problems was seen in the oral, pharyngeal and esophageal phase. In fact, greater percentage of children had problems in the oral phase compared to the other two phases of swallow. These findings are similar to the findings obtained in the VHRG, except for the findings related to vomiting after feeding and nasal regurgitation. Greater percentage of children in the clinical group experienced vomiting after feeding and nasal regurgitation compared to the VHRG. This could be attributed to the underlying differences in the medical conditions experienced by the two groups. Majority of the children in the clinical group had a nervous system damage leading to cerebral palsy. GERD is one of the serious problems which can be present in 70-75% of

children with CP. CP children with GERD present with various feeding problems. They can have recurrent vomiting and chest infections, reactive airway disease such as nocturnal asthma, choking attacks, anaemia and wheezing which leads to poor growth and nutrition (Gangil, Patwari, Bajaj, Kashyap, & Anand, 2001). Soft palate weakness reported is also seen in cerebral palsy which could lead to nasal regurgitation (van den Engel-Hoek et al., 2015).

Additionally, based on the second assessment, an attempt was made to categorize the infants into those with feeding and swallowing difficulties (dysphagia) and those without any difficulties. It was seen that 5 participants (7.5%) from the HRG and 16 participants (48.5%) from the VHRG continued to have feeding and swallowing difficulties (dysphagia). All the participants from the TG, 62 participants (92.5%) from HRG and 17 (51.5%) from the VHRG had adequate feeding and swallowing abilities and did not have dysphagia. Table 4.10 shows the status of feeding/swallow function of all the participants after the second assessment. The overall NDST scores of those who were identified with “adequate feeding and swallowing abilities” and those identified with “inadequate feeding and swallowing abilities (dysphagia)” were compared using Mann-Whitney test to check the presence of any significant difference. The results revealed that there was a very high significant difference ( $|z|=6.05$ ,  $p < 0.001$ ) between these neonates.

Table 4.10

*Feeding/swallow status of all participants after the second assessment using NDST*

<b>Groups</b> (N = 178)	<b>Feeding/swallow status</b>	
	<b>Adequate ability (%)</b>	<b>Inadequate ability (%)</b>
Typical Group (N=75)	75 (100)	0 (0)
High Risk Group(N=67)	62 (92.5)	5 (7.5)
Very High-Risk Group(N=36)	17 (51.5)	16 (48.5)

Subsequently, the NDST scores of the neonates categorized as “inadequate feeding and swallowing abilities (dysphagia)”, based on the results of the second assessment, were compared with the NDST scores of the clinical group of children whose problems were assessed retrospectively using the Mann-Whitney test. The results revealed that there was no significant difference between these two groups ( $|z|=0.28$ ,  $p>0.05$ ). This indicated that both these groups had exhibited feeding and/or swallowing difficulties. This indirectly reflects the high sensitivity of the tool.

### **Section III: Sensitivity and Specificity of NDST**

The overall sensitivity of NDST ranged from 66.67% to 100% when identifying the feeding and swallowing difficulties of children retrospectively during their neonatal period. Similarly, the overall specificity of NDST ranged from 50.85% to 69.77% when identifying the children without feeding and swallowing difficulties. Further, the average sensitivity and specificity of NDST were 89.12% and 55.59% respectively. Table 4.11 shows the sensitivity and specificity of NDST.

This finding can be interpreted as a high sensitivity (89.12%) and moderate specificity (55.59%) of the tool. The high sensitivity values indicated that NDST had higher probability of detecting the presence or absence of feeding and swallowing difficulties. The moderate specificity could be due to increased false positive responses (44.41%).

When the sensitivity and specificity of NDST were compared with Neonatal Feeding Assessment Scale (NFAS, Viviers et al., 2016), which is a similar tool that assesses feeding in neonates, it was found that NFAS had higher sensitivity (100%) and specificity (78.6%). This difference could be attributed to the nature of the tool i.e., NFAS being a comprehensive clinical feeding assessment instrument, whereas NDST is a screening tool. Also, the smaller sample size used in the standardization of NFAS (n=20) and variations in the participant sample characteristics could have contributed to the differences.

The NDST's sensitivity and specificity was also compared to the Dysphagia Screening Test for Preterm Infants (DST-PI, Leo & Seo, 2017), which again had higher sensitivity (96.6%) and specificity (76.9%). This variation could be due to the study population considered, i.e. only premature neonates were included in the standardization of DST-PI, while neonates with all types of risk factors including prematurity was included in the present study.

Limited information was available on the sensitivity and specificity of other similar feeding and swallowing assessment tools in neonates and infants such as Early Feeding skills (Da Costa, Van Den Engel-Hoek & Bos, 2008; Pados et al., 2016). Hence a comparison with the tool developed in the present study was not feasible.

Table 4.11

*Sensitivity and specificity values of NDST*

<b>Q.Code</b>	<b>Sensitivity (%)</b>	<b>Specificity (%)</b>
Q1	100	52.63
Q2	100	53.57
Q3	100	66.67
Q4	100	69.77
Q5	100	69.77
Q6	100	66.67
Q7	100	56.60
Q8	66.67	50.88
Q9	100	50.85
Q10	66.67	51.85
Q11	100	50.85
Q12	91.67	60.42
Q13	100	53.57
Q14	100	60
Q15	100	52.63
Q16	87.50	63.64
Q17	91.67	60.42
Average	89.12	55.69

#### **Section IV: Inter-rater reliability of NDST**

In order to verify the inter-rater reliability of NDST, data was collected from 18 participants (10% of the sample) by two independent investigators. Inter-rater reliability of NDST was determined using Cohen's kappa coefficient and interpreted using the guidelines given by Parikh et al. (2008). Generally, a Kappa value of greater than 0.6 is considered to have good reliability.

The results indicated a perfect agreement ( $k=1$ ) for all the questions except Q4 which had a good agreement ( $k=0.77$ ). Further, the overall inter-rater reliability also indicated a perfect agreement ( $k=0.99$ ) between the two independent raters. Therefore, the findings indicated a very good inter-rater reliability of the NDST. This clearly shows that all clinicians using the NDST on neonates are likely to obtain the same results. Hence, a rigorous guidance and training on NDST administration may not be required to support a clinician in achieving consistent results when administering the NDST.

In addition, NDST had a very good inter-rater reliability when compared with other commonly used instruments investigating feeding and swallowing components such as NOMAS (Palmer et al., 1993), SOMAS (Reilly et al., 2000) and NFAS (Viviers et al., 2016). Palmer et al., (1993) had not assessed the inter-rater reliability of NOMAS at the time publishing their final scale. Later, Da Costa and Van der Schans in 2008 reported a moderate to substantial agreement (Kappa = 0.40 to 0.65) on evaluating the inter-rater reliability of NOMAS. The SOMAS had an excellent agreement beyond chance on the inter-rater reliability testing with the Kappa value of more than 0.75 on a sample of 10 children (Reilly et al., 2000). However, these two tools are focussed more towards assessing the oromotor aspects associated with feeding. The NFAS (Viviers et al., 2016) showed a substantial agreement beyond chance with the Kappa value of

0.74 in the inter-rater reliability testing. However, the authors of EFS (Pados et al., 2016) reported that reliability between and within raters (intra- and inter-rater reliability) was found to be consistent and acceptable, though no data was provided to support this statement (Da Costa & Van der Schans, 2008).

## **Section V: Critical test items of NDST**

The scores on the first administration of NDST was considered for identifying the critical test items of the tool. This was because of the nature of the participant group and the method adopted for collecting the data, i.e., the NDST was administered by the investigator by obtaining the relevant information from the mothers of the neonates. Further, the tool was developed with the intention of identifying dysphagia in neonates. The critical test items of the NDST were defined by the criteria developed by the investigators of the present study. In this criterion, midpoint of the range of “yes” responses (in percentage) of all questions in the VHRG was considered to be the minimum cut-off score to be marked as critical test item of NDST. The reason for considering only the VHRG group to mark the critical test item was due to the sample characteristics i.e., only the VHRG contained neonates with very high-risk factors that can lead dysphagia or feeding and swallowing difficulties (Jadcherla, 2016). Table 4.12 shows the percentage of ‘yes’ responses of the participants in the first administration of NDST across the groups.

The percentage of “yes” responses varied from 0% to 13.3% in TG, 0% to 26.9% in HRG and 0% to 42.4% in VHRG. The midpoint of these ranges was 6.65% in TG, 13.45% in HRG and 21.2% in VHRG. Based on the criteria to mark the critical test items of NDST, only the VHRG group was considered and the midpoint was 21.2%. Consequently, the questions with  $\geq$  21.2% of “yes” response across all groups were considered as critical test items of NDST.

Consequently, the critical test items of NDST were Q4 (staying fixed to the breast), Q5 (delayed initiation of sucking), Q6 (requiring stimulation to initiate sucking), Q9 (breathing difficulty during feeding), Q10 (short feeding duration), Q12 (anterior spillage of fluid), Q14 (coughing/choking/gagging/tearing/turning blue) and Q16 (frequent hiccups after feeding). Since Q6 indirectly indicated delayed initiation of sucking, the same was not included under the critical test items.

Table 4.12

*Percentage of 'yes' responses obtained in the first administration of NDST across the groups*

<b>Q.</b>	<b>% of yes in TG</b>	<b>% of yes in HRG</b>	<b>% of yes in VHRG</b>
<b>Code</b>	<b>(N = 75)</b>	<b>(N = 67)</b>	<b>(N = 36)</b>
Q1	1.3	1.5	3
Q2	0	7.5	9.1
Q3	9.3	13.4	18.2
<b>Q4*</b>	<b>9.3</b>	<b>16.4</b>	<b>27.3*</b>
<b>Q5*</b>	<b>6.7</b>	<b>17.9</b>	<b>21.2*</b>
<b>Q6*</b>	<b>8</b>	<b>26.9*</b>	<b>18.2</b>
Q7	1.3	3	18.2
Q8	1.3	3	0



<b>Q9*</b>	<b>0</b>	<b>1.5</b>	<b>21.2*</b>
<b>Q10*</b>	<b>6.7</b>	<b>7.5</b>	<b>30.3*</b>
Q11	1.3	3	0
<b>Q12*</b>	<b>2.7</b>	<b>10.4</b>	<b>30.3*</b>
Q13	1.3	1.5	6.1
<b>Q14*</b>	<b>2.7</b>	<b>11.9</b>	<b>42.4*</b>
Q15	1.3	0	3.0
<b>Q16*</b>	<b>13.3</b>	<b>22.4*</b>	<b>1.2</b>
Q17	12	16.4	18.2

Note\*: \*Critical test items; TG-Typical group; HRG-High risk group; VHRG-Very high-risk group.

## Section VI: Establishment of interpretation algorithm for NDST score

Of the different sections of NDST, only section E comprises of questions which objectively assess the feeding and swallowing abilities using a scoring system (a score of 1 for a ‘yes’ response and a score of 0 for a ‘no’ response). All other sections help to describe and profile the aspects that are relevant to feeding and swallowing. To find the total score on the NDST, one has to add the number of “yes” responses obtained on the questions in the section E. Table 4.13 shows the distribution of overall scores of NDST across the groups.

The maximum overall score of the participants in the TG, HRG and VHRG was “4”, “8” and “14” respectively. In order to facilitate the interpretation of the overall scores of NDST, the following algorithm was developed based on the maximum overall score of the participants across the groups. The maximum overall score of the participants in the TG, HRG and VHRG was considered as upper cut off of the range to label them as low, moderate and high risk for dysphagia respectively. Consequently, the overall scores of NDST between 0-4 could be considered as “low risk” for dysphagia; the scores ranging between 5-8 could be considered as “moderate risk” for dysphagia and the scores ranging between 9-14 could be considered as “high risk” for dysphagia. Further, the scores greater than 14 could be considered as “very high risk” for dysphagia.

Table 4.13

*Distribution of overall scores of NDST across the groups*

<b>Overall score</b>	<b>TG (%)</b>	<b>HRG (%)</b>	<b>VHRG (%)</b>
<b>(N = 178)</b>	<b>(N = 75)</b>	<b>(N = 67)</b>	<b>(N = 36)</b>
0	68	37.3	12.5
1	12	17.9	3.1
2	8	13.3	12.5
3	5.3	6.0	0
4	6.7	6.0	12.5

5	0	9.0	0
6	0	6.0	12.5
7	0	3	3.1
8	0	1.5	12.5
9	0	0	3.1
10	0	0	12.5
11	0	0	3.1
12	0	0	3.1
13	0	0	6.4
14	0	0	3.1
15	0	0	0
16	0	0	0
17	0	0	0

Note\*: TG-Typical group; HRG-High risk group; VHRG-Very high-risk group

Overall, the results revealed that the feeding and swallowing difficulties were significantly higher in the neonates in the VHRG, followed by the infants in the HRG on the first and second assessment using the NDST. There was a significant difference between the VHRG

and HRG as well. On comparison of the median scores of first and second administration of NDST across groups, it was observed that the scores had reduced considerably for the participants in the TG and HRG, however, there was no reduction in the median scores in the VHRG. Further statistical test revealed a very high significant difference between both the assessments for TG and HRG. However, for VHRG, no significant difference was observed between first and second assessment, which indicated that the feeding and swallowing difficulties in the VHRG persisted.

In the third assessment again, the neonates in the VHRG obtained significantly higher scores than the HRG. The development of infants in HRG and VHRG was also assessed using “Remember & Care checklist” at the end of four months. It was seen that greater number of infants in the VHRG had delayed development compared to the infants in the HRG in hearing, speech and language and motor domains.

The NDST was administered on another clinical group (children with a confirmed diagnosis of oro-pharyngeal dysphagia in the age range of 0.6 to 4 yrs) and an age and gender matched typical group (typically developing children) and the scores were compared across the groups. These findings indicated that the clinical group had significantly greater feeding difficulties than the typical group. Further, NDST had high sensitivity (89.12%), moderate specificity (55.59%) and a very good inter-rater reliability of the NDST.

## Chapter V

### Summary and Conclusions

Oral feeding is the most complex and vital sensori-motor process in the new-borns. Feeding difficulties can occur frequently in neonates due to associated medical conditions such as prematurity, low birth weight, cardiopulmonary diseases and neurological disorders (Jadcherla, 2016). Early identification of feeding and swallowing problems in newborns would facilitate the early intervention, thereby contributing to a better quality of life in the neonate and the family members.

The present study aimed at developing a screening tool for the detection of dysphagia in neonates and standardizing it by establishing the validity and reliability. The tool that was developed was referred to as Neonatal Dysphagia Screening Tool (NDST). The specific objectives of the study were to compare the responses obtained using the NDST across typical, high risk and very high-risk neonates in the age range of 1-5 days, to establish the sensitivity and specificity of the developed tool by administering it retrospectively on a known paediatric population with and without dysphagia in the age range of 0.6 to 4 years, to identify the critical test items of the developed tool and to establish its scoring and interpretation algorithm.

The NDST was validated for its contents by a multidisciplinary group of professionals and was subjected to a pilot study, following which the final version was frozen. The final version of NDST was administered on 178 mothers of neonates in the age range of 1-5 days (99 males and 79 females), who were divided into three groups based on the medical / birth history such as typical group (TG-75 participants), high risk group (HRG-67 participants) and very high risk group (VHRG-36 participants), as a part of assessment of clinical validity. The risk factors

in the HRG included thyroidism, epilepsy and chicken pox (viral/bacterial infections) in the pre-natal period, foetal distress, aspiration of amniotic fluid, moderate to late prematurity (gestational age: 32-37 weeks), low birth weight (LBW-1500 to 2500 g), high birth weight (HBW->4000g), delayed birth cry and moderate birth asphyxia (APGAR scores:4-6) in the peri-natal period and neonatal jaundice, hypoglycaemia (low sugar level), viral/bacterial infections, and high fever in the post- natal period and this group was labelled as high risk group (HRG). The risk factors in the VHRG included extremely low birth weight (ELBW-<1500g), extreme prematurity (gestational age: 28-32 weeks), serious birth asphyxia (APGAR scores: 0-3), congenital anomalies, respiratory distress syndrome (RDS), hypoxic-ischemic encephalopathy (HIE) and convulsion.

All the participants in the three groups, irrespective of presence or absence of feeding and swallowing difficulties, were followed up at the end of one month to assess if there were any changes in the feeding and swallowing functions over time. A third assessment was also performed at the end of four months only for the HRG and VHRG. This was done for 57 participants only (40 from HRG and 17 from VHRG). Additionally, a screening checklist titled “Remember & Care” (developed at the Department of Prevention of Communication Disorders) which screens for motor, hearing, speech and language milestones was administered to assess, if any of the infants exhibited any developmental delays.

In order to establish the sensitivity and specificity effectiveness of NDST, the tool was administered on a different group of participants in the age range of 0.6 to 4 years with (N=30) and without dysphagia (N=30). The NDST was administered on 10% (18) of the young neonates in the age range of 1 to 5 days by two other qualified speech-language pathologists (SLP’s) for the purpose of analysing its inter-rater reliability.

The response of the mothers of the neonates on each question was converted into a score based on the rating scale. These were added up to obtain a total score. This was done on the first, second and third administration of NDST. The total score was also obtained from the clinical and control group which were additionally included in the study. The total scores were averaged and obtained for all the groups.

The obtained data was analysed using SPSS (version 20) statistical software. Descriptive statistics was used to obtain mean, median and standard deviation of overall scores obtained on the NDST. Mann Whitney U test was used to check for any significant differences between the TG, HRG and VHRG that existed for the first (within 5 days of birth) and second administration (at the end of one month) of NDST. Kruskal-Wallis test was used to test the presence of any significant difference across all the three groups (TG, HRG and VHRG) based on the total score for all the three administration of NDST. Wilcoxon test was performed to identify any significant differences between first (within 5 days of birth) and second (at the end of one month) administration of NDST across the groups. Chi-square test was performed to measure the level of significant association between mother's response and the groups on the NDST. For the inter-rater reliability, Cohen's kappa coefficient was used to identify the level of agreement.

The results revealed that the feeding and swallowing difficulties were significantly higher in the neonates in the VHRG, followed by the infants in the HRG on the first and second assessment using the NDST. There was a significant difference between the VHRG and HRG as well. On examining each test item of the NDST on all the groups during the first assessment, neonates in the VHRG demonstrated feeding and swallowing difficulties on 10 questions. It was seen that maximum percentage of them had coughing / gagging / choking / tearing or watery eyes/ turning to blue during or immediately after feeding, anterior spillage of fluid and shorter

feeding span, which were greater than that seen in the HRG and TG. The other most frequently observed feeding and swallowing difficulties in the VHRG included difficulty in staying fixed to the breast, taking longer time to suck, breathing difficulties during and after feeding, which were again greater than that seen in HRG and TG. These findings indicated that they had difficulties with the pharyngeal and the oral phase of swallow.

Second assessment using NDST at the end of first month showed negligible percentage of issues in TG (4%, Q16 and 1.3%, Q14). However, in the HRG, 10.4% neonates had difficulty with Q14 & 16. Only a negligible percentage of them had difficulties on Q 4,9,10,12,13,14,16&17. In VHRG, 60.6% of the neonates had frequent coughing/choking/gagging/tearing or watery eyes/turning into blue during or immediately after feeding (Q14); 48.5% had frequent hiccups after feeding (Q16); 45.5% reported leak of fluid from their babies mouth (Q12), 27.3% and 24.3% reported that their babies had difficulty in staying fixed to the breast and delayed sucking after latching (Q4 & Q5) and 21.1% had difficulty in initiating a swallow and vomited after feeding. All the other difficulties were reported by less than 10% of the neonates in the VHRG.

On comparison of the median scores of first and second administration of NDST across groups, it was observed that the scores had reduced considerably for the participants in the TG and HRG, however, there was no reduction in the median scores in the VHRG. Further statistical test revealed a very high significant difference between both the assessments for TG and HRG. However, for VHRG, no significant difference was observed, which indicated that the feeding and swallowing difficulties in the VHRG persisted.



In the third assessment again, the neonates in the VHRG obtained significantly higher scores than the HRG. The development of infants in HRG and VHRG was also assessed using “Remember & Care checklist” at the end of four months. It was seen that greater number of infants in the VHRG had delayed development compared to the infants in the HRG in hearing, speech and language and motor domains.

The NDST was administered on another clinical group (children with a confirmed diagnosis of oro-pharyngeal dysphagia in the age range of 0.6 to 4 yrs) and an age and gender matched typical group (typically developing children) and the scores were compared across the groups. These findings indicated that the clinical group had significantly greater feeding difficulties than the typical group. Further, NDST had high sensitivity (89.12%), moderate specificity (55.59%) and a very good inter-rater reliability of the NDST.

Thus the outcome of the study is a highly sensitive, valid and reliable screening tool to identify dysphagia in neonates, which can be used by speech-language pathologists (SLPs). The administration of this does not require any rigorous training. It is quickly interpretable, time and cost-effective. It includes all the relevant areas of neonatal feeding prominently, which in turn facilitates the early identification of dysphagia in high risk neonates. The test items of NDST reflect the wide array of skills and components, which forms the foundation of neonatal feeding behaviour and therefore is a comprehensive instrument. In addition, the critical items of the NDST have been identified and interpretation algorithm also has been provided, which helps the SLPs in quantifying the extent of dysphagia in the neonates by providing a score. Based on this, one can categorize the infants into low, moderate, high and very high risk for dysphagia. Also, this quantitative scores of NDST obtained will strengthen the clinical findings made by the SLPs. Therefore, the NDST will be a very useful tool for routine use in newborn screening programs.

Further, the administration of NDST is by eliciting relevant information from the mothers. Mothers contribute greatly to feeding assessment by providing information about their infant, and their experience and feelings surrounding the feeding. This kind of a family-centered developmentally supportive approach relates to current evidence in the field of neonatal dysphagia (Lau & Smith, 2011; Thoyre et al., 2013).

However, there are few limitations in the present study. The sensitivity and specificity of NDST was established by administering it on an older group of children diagnosed with dysphagia. Instead, clinical validity of NDST could have been derived by comparing the quantitative NDST score of the neonates with the objective evaluation of swallowing for the same group of infants. The tool is designed for use by SLPs. However, in order to screen every newborn for feeding and swallowing issues, it is important that the nurses also administer this tool, considering the fact that not all hospitals have SLPs in place. Future studies can be undertaken to test the reliability of the tool when administered by nurses as against the SLPs.

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**Appendix**  
**Neonatal Dysphagia Screening Tool (NDST)**

**Section A: Demographic data**

Name: \_\_\_\_\_ Hospital Name: \_\_\_\_\_  
 Gender: \_\_\_\_\_ Date of birth: \_\_\_\_\_  
 Chronological Age: \_\_\_\_\_ Birth order: \_\_\_\_\_  
 Languages known: \_\_\_\_\_ Address: \_\_\_\_\_  
 Phone number: \_\_\_\_\_  
 Landline: \_\_\_\_\_  
 Mobile: \_\_\_\_\_  
 Email-id: \_\_\_\_\_

	<b>Maternal details</b>	<b>Paternal details</b>
Name		
Age		
Education		
Occupation		
Family Income		
	Overall SES:	

**Section B: Medical History**

**i) Pre-natal history:**

Type of conception: Natural/In vitro  
 Family history of disability: Present/absent  
 Other issues: Viral/bacterial infections (specify)/Thyroidism/Epilepsy

**ii) Peri-natal history:**

Type of delivery: Normal/ LSCS/ Breech/ Instrumental  
 Gestational age: Extremely pre-term (28-32 weeks)/ moderate to late pre-term(32-37weeks)/ Full term(38-41weeks) /Post mature (>48 weeks)  
 Corrected Age:  
 Birth cry: Immediate / Delayed by \_\_\_\_\_minutes  
 Colour at birth:  
 Birth weight: >4000g - HBW, 2500-3999g - NBW, 1500-2499g-LBW, 1000-1499g-VLBW, <999g- ELBW (Southgate & Pittard, 2001)

Birth Asphyxia: 0-3: Serious asphyxia, 4-6: Moderate asphyxia, 7-10: Normal (Based on APGAR score, Rossetti, 2001)

Other risk factors: Foetal Distress/Cord prolapse/Aspiration of amniotic fluid

**iii) Post-natal history:**

Congenital anomalies (including craniofacial anomalies) (specify)/ Neonatal jaundice/High fever/Viral/bacterial infections/Convulsions/Respiratory distress syndrome/Hypoxic-Ischemic events/hypoglycaemia

Medical treatment, if any:

NICU stay: Yes/No

Ventilator usage: Yes/No

other Remarks:

**Section C: Physical/Physiological functioning**

i) Neonatal state: Calm & Alert/ Lethargic

**ii) Neonatal Oral Reflexes**

Rooting reflex (Stroke the cheek and the child will turn toward the cheek that was stroked)	Present	Absent
Suck-Swallow reflex (Touch the lips, the mouth opens and suckling movements begin)	Present	Absent
Tongue thrust reflex (When the lips are touched, the child's tongue extends out of the mouth)	Present	Absent
Gag reflex (Elicited by touching the posterior pharyngeal wall)	Present	Absent
Phasic bite reflex (Apply light pressure to the gums results in early munching patterns)	Present	Absent
Transverse tongue reflex (Touch on either side of the tongue results in tongue moving to the side of touch)	Present	Absent

**iii) Oro-Motor Examination**

Oral structure	Structure	Function
Upper lip		
Lower lip		
Tongue		
Hard palate		
Soft palate		
Jaw		

\*Must look for lip seal, tongue thrust, temporo-mandibular joint movement and any other structural abnormalities.

iv) **Drooling:** Present/absent

**Section D: Feeding history**

- i) Do you feel that your child has feeding/swallowing difficulty? Yes / No
- ii) What is the feeding method used?  
Naso-gastric tube feeding/Gastrostomy tube feeding/Breast feeding/Bottle feeding  
Paladai feeding/Spoon feeding/Others
- iii) Do you use any special feeding aids? Yes (specify)/No
- iv) What is the total feeding duration?  
<10 minutes/10-20 minutes/21-30 minutes/>30minutes (specify)
- v) How often do you feed your baby?  
Every half an hour/Every hour/Every 2 hours/ Others (specify)
- vi) Does your child make eye contact during feeding? Yes/No
- vii) Does your child indicate hunger? Yes (specify) /No  
Crying/Waking up and acting restless/Sucking on hand/Smacking lips/Opening and closing mouth/Rooting around on the chest of the person who is carrying/ Others
- viii) Does your child indicate satiety? Yes (specify) /No  
Decreasing or stopping sucking/Pushing nipple away from mouth/Falling asleep/  
Crying and turning head away from nipple/Showing increased interest in surroundings rather than drinking/Others
- ix) Are you satisfied with the quantity of milk/food intake? Yes/No
- x) Are you happy with the baby’s current weight? Yes/No
- xi) Does your child seem satisfied/calm after feeding? (Rest happily in your arms or falls asleep) Yes/No

**Section E: Feeding and swallowing assessment**

**Scoring:** “Yes” indicates the presence of difficulties and “no” indicates the absence of difficulties. Assign a value of 1 for a ‘yes’ response and a value of 0 for a ‘no’ response.

Sl.No	Questions	Responses	
		Yes	No
1.	Does your child have difficulty in turning head towards you during feeding?	Yes	No
2.	Does your child have difficulty in searching for the nipple or opening the mouth in response to the breast/bottle/spoon during feeding?	Yes	No
3.	Does your child have difficulty in latching on to the breast?	Yes	No
4.	<b>Does your child have any difficulty in staying fixed to the breast actively?</b>	Yes	No
5.	<b>Does your child take long time to suck immediately after latching?</b>	Yes	No



6.	<b>Does your child need any sort of stimulation to initiate a suck? (If yes, Specify the type of stimulation used _____)</b>	Yes	No
7.	Does your child keep the milk in the mouth without swallowing?	Yes	No
8.	Does your child refuse to drink milk/take feeds?	Yes	No
9	<b>Does your child have any breathing difficulty/ loud breathing noises during or immediately after feeding?</b>	Yes	No
10	<b>Does your child have short period of feeding (less than 5 minutes)?</b>	Yes	No
11.	Does your child have prolonged period of feeding (more than 30 minutes)?	Yes	No
12.	<b>Does the fluid (milk) leak from his/her mouth while feeding?</b>	Yes	No
13.	Does milk come out of the nose during or immediately after feeding?	Yes	No
14.	<b>Does your child have frequent coughing/choking/gagging/tearing/turning into blue during or immediately after feeding?</b>	Yes	No
15.	Does your child have wet/gurgly voice/cry after feeding?	Yes	No
16.	<b>Does your child have frequent hiccups after feeding?</b>	Yes	No
17.	Does your child have vomiting after feeding?	Yes	No
<b>Total score</b>			

**\*Critical items are in bold font**

**Interpretation of total score of NDST:** 0-4 = Low risk for dysphagia; 5-8 = Moderate risk for dysphagia; 9-14 = High risk for dysphagia and <14 = Very high risk for dysphagia

**Impression:**

**Recommendations:**

Name of the Examiner:

Signature:

Date: