

**AN INVESTIGATION OF THE FEEDING PROBLEMS IN
CHILDREN WITH DOWN SYNDROME**

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CERTIFICATE

This is to certify that this dissertation entitled “**An Investigation of the Feeding Problems in Children with Down syndrome**” is a bonafide work submitted in part fulfillment for the Degree of Master of Science (Speech-Language Pathology) of the student (Registration No.: 13SLP013). This has been carried out under the guidance of a faculty of this institute and has not been submitted earlier to any of the University for the award of any Diploma or Degree.

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This is to certify that this dissertation entitled “**An Investigation of the Feeding Problems in Children with Down syndrome**” has been prepared under my supervision and guidance. It is also certified that this has not been submitted earlier to any of the University for the award of any Diploma or Degree.

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DECLARATION

This is to certify that this dissertation entitled “**An Investigation of the Feeding Problems in Children with Down syndrome**” is the result of my own study under the guidance of Dr. Swapna. N., Reader in Speech Pathology, Department of Speech-Language Pathology, All India Institute of Speech and Hearing, Mysore, and has not been submitted earlier in other University for the award of any Diploma or Degree.

Mysore
May 2015

Register No.: 13SLP013

DEDICATED TO

DAD & MOM

&

TO MY REMARKABLE

GUIDE

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Chapter 1

Introduction

Feeding is a sensorimotor skill that matures during the first two years of life. It is a skill present from early infancy which provides nutrition for normal growth and development. The satisfaction of hunger and maintenance of homeostasis is achieved through the feeding process. Feeding also provides opportunities for sensory and motor stimulation, mother-child bonding and oro-motor skill development (Kummer, 2008). It is a highly complex process with developmental stages based on the neurological maturation and experiential learning of the child. However, feeding, unlike other sensorimotor skills, is heavily dependent on internal incentives or motivation to initiate ingestion such as hunger cry from birth on, which is essential for survival of the newborn (Lewis, 1982; Morris, 1982; Sheppard & Mysak, 1984).

Feeding involves the neurological control of refined, synchronised and coordinated muscle movements. Consequently several factors might influence feeding. First, adequate neurological function and intact swallowing reflex is necessary for the child to successfully manage solid and liquid boluses orally. Secondly, adequate muscle tone, control, and coordination are essential. Children who self-feed must be able to sit in an appropriate position, gather and hold the food with fingers, use a spoon or cup and move it to their mouth successfully. A third factor is the oral-motor skill and function. Adequate oral motor development which follows a stepwise progression from suckling, the most primitive oral motor pattern, to the more complex oral-motor milestones of

suck, munch and chew is essential (Bosma, 1986). The other factors that influence this process are the anatomical, environmental, cultural and social factors.

The oral cavity, pharynx, and esophagus are three anatomical regions that play an important role in feeding. They can function separately but, in swallowing they effectively integrate and coordinate their functions through a neuronal network. Normal swallowing mechanism includes an integrated independent group of complex feeding behaviors emerging from the interactions of cranial nerves of the brainstem, which is governed by the neural regulatory mechanisms in the medulla oblongata, as well as in higher cortical and subcortical structures. Such sensory guided discriminatory feeding and sensory-cued, stereotyped swallowing mechanism of our body is described in four phases: The oral preparatory phase, the oral phase, the pharyngeal phase and the esophageal phase (Logemann, 1998). During the oral preparatory phase, both voluntary control and reflexive components are integrated with feeding and chewing. In this phase food is manipulated in the mouth in an attempt to reduce it to a consistency ready for swallow. This action requires sufficient lip closure, tongue and jaw movement. In the oral phase, the prepared food is pushed into the oropharynx and then into the hypopharynx until the pharyngeal swallow is triggered. The tongue is the key structure for this phase. In the pharyngeal phase, the pharyngeal swallow is triggered and the bolus is moved through the pharynx into the esophagus along with a few mechanical adjustments made to make sure that the airway and nasal cavities are protected from the food particles, so that it will not lead to aspiration or nasal regurgitation. During the esophageal phase, the upper esophageal sphincter opens and the esophageal peristalsis carries the bolus through the cervical and thoracic esophagus and into the stomach via the

lower esophageal sphincter. The duration and characteristics of each of these phases depend on the type and volume of food being swallowed and the voluntary control exerted over it (Kahrilas & Logemann, 1993; Kahrilas, Lin, Chen, & Logemann, 1996).

In normal infants, feeding skills mature in a sequence that parallels with the development of other motor skills. The development of feeding skills is an extremely complex process. In most children, the individual acts during feeding and swallowing occur normally enabling them to take in food with ease. However, in some children the problems in feeding can arise due to problems in any of the phases, which in turn can lead to dehydration, malnutrition and respiratory symptoms. These infants and children experiencing feeding difficulties would demonstrate various signs and symptoms such as difficulty in breast feeding, difficulty in coordinating breathing with eating and drinking, difficulty in sustaining oral feeding in order to maintain adequate caloric intake, difficulty in sucking, biting, chewing and swallowing, difficulty in accepting different textures of food, partial to total food refusal, delay in self-feeding, or coughing or gagging during meals. The limited experiences with oral intake related to the medical or physical conditions of the child, as well as other variables such as prematurity, often result in a failure to successfully thrive and develop the normal oral motor skills. Over the past 35-40 years pediatric feeding problems have been recognized in the literature by various authors (Logan & Bosma, 1967; Illingworth, 1969).

Any anatomical and/or physiological defects in the structures can cause feeding problems in all the phases, as these organs work in close coordination with one another. If any one phase is functionally impaired, the chances of other phases being affected will also increase. This can cause feeding to be unpleasant, detestable, negative, or even

excruciating. Etiological factors contributing to feeding problems are often multifactorial which can vary from illness, injury, metabolic diseases, sensory defects, tumors, infections, neurological defects, structural anomalies, gastrointestinal disorders as well as genetic conditions. Feeding problems are common in children with different genetic syndromes due to the complex interactions between medical, anatomical, physiological, and behavioral factors. Some of the genetic conditions frequently resulting in feeding issues are CHARGE syndrome, Noonan syndrome, Pierre Robin syndrome, Velocardiofacial syndrome, Williams syndrome, DiGeorge syndrome, Cornelia de Lange syndrome, Klippel Feil syndrome, Sotos syndrome, Prader Willi syndrome and Down syndrome (Samuels & Chadwick, 2006).

Down syndrome is one of the genetic conditions in which the feeding problems seen have been documented in the literature. Van Dyke, Peterson and Hoffman in 1990 observed that many individuals with Down syndrome have potential feeding problems as a consequence of oral, anatomical and structural anomalies. Several studies of oral feeding in children with Down syndrome, particularly young children have shown a 50-80 % frequency of feeding problems (Pipes & Holm, 1980; Van Dyke et al., 1990). Down syndrome occurs due to a chromosomal anomaly and it is one of the most common genetic syndromes, occurring in 1 of 800 to 1,000 live births (Baird & Sadovnick, 1989). Children with Down syndrome have three copies of the genes on chromosome 21, rather than the usual two resulting in trisomy 21, which is seen in approximately 95 percent of the cases (Cooley, 1993). The anatomical and physiological characteristics such as neuromotor coordination impairments, craniofacial and structural abnormalities and dysfunctions seen in these individuals have direct and indirect consequences on feeding

abilities (Cooper-Brown, Copeland, Dailey, Downey, Petersen, Stimson, & Van Dyke, 2008). These different structural and anatomical aspects of Down syndrome frequently interfere with the acquisition of effective oral-motor skills and can result in these children developing potential feeding problems and swallowing dysfunction (Cohen, Winner, Schwartz, & Skalar, 1961).

Longitudinal studies by researchers (Calvert, Vivian, & Calvert, 1976; Pipes & Holm, 1980; Cullen, Cronk, Pueschel, Schnell, & Reed, 1981; Aumonier & Cunningham, 1983; Spender, Stein, Dennis, Reilly, Percy, & Cave, 1996) addressing the different feeding and oromotor problems of subjects with Down syndrome report problems with breast and bottle feeding, a poor or delayed suckling ability, problems with mastication, drooling, a tendency to allow the mouth to hang open at rest, a protruded tongue posture persisting beyond the age of 2 years, a delay in dental eruption, presence of a malocclusion, slow development of the ability to manipulate food in fingers and in the use of feeding utensils; the failure to progress through a normal sequence of food textures and the refusal of certain foods, particularly those of a hard texture; behavioral problems such as refusal to swallow, spitting out of food, or retention of food or utensils in mouth. The oral dysmorphology due to malocclusion can significantly impact feeding process due to the poor contact of the upper and lower arches of teeth, which can prevent adequate chewing and grinding of foods. Along with feeding and oral motor problems, frequent dental anomalies such as periodontal disease, tooth loss and severe bruxism, increases the risk of developing feeding problems in them. Several comorbidities like chronic illnesses such as anatomical abnormalities, congenital heart disease, dental disease, pulmonary complications, esophageal, stomach, and bowel dysmotility, and

gastroesophageal reflux disease are common problems leading to poor feeding and impoverished growth in Down syndrome (Cronk, Crocker, Pueschel, Shea, Zackai, Pickens, & Reed, 1988).

Further most infants with Down syndrome have systemic low tone which affects the muscles of the tongue, lips, cheeks, face, jaw, pharynx and larynx. This can lead to inadequate strength, speed and range of muscle movements which are vital for functions of sucking, chewing, biting, and swallowing. Parents commonly report of the following concerns of weak suck and lip closure, difficulties in biting and chewing, and uncoordinated swallow leading to choking and gagging (Van Dyke et al., 1990). Mitchell, Call, and Kelly in 2003 reported that obstructive sleep apnea and laryngomalacia were found to be the most common disorders in children with Down syndrome which were likely to have a significant impact on the coordination of breathing and swallowing rhythm.

Several studies have been carried out to identify the different feeding problems in children with Down syndrome. These problems could affect any of the four phases of swallowing. Studies on infants with Down syndrome reported that the oral complications seen in infants can cause it to be extremely challenging to breastfeed (Cunningham, 1996; Skallerup, 2009). Torfs and Christianson (1998) studied anomalies at birth in 2.5 million control infants in which 2,894 infants were those with Down syndrome and found a higher incidence of hypotonia, cardiac defects, digestive system problems, eye cataract, respiratory system defects, genital differences, anomalies of the extremities, urinary system problems, hydrocephalus, hernia, and cleft palate in infants with Down syndrome which interfere with breast feeding. In 2003, Pisacane, Toscano, Pirri, Continisio, Andria,

and Zoli studied the prevalence and duration of breastfeeding among infants with Down syndrome and found breast feeding to be significantly lower with a mean duration of breastfeeding days being 54 days in infants with Down syndrome as compared to 164 days in infants without Down syndrome.

Another feeding examination was carried out by Lewis and Kritzinger in 2004 on 0-3 month's old infants with Down syndrome. Twenty infants with Down syndrome were assessed via parental questionnaire. The feeding problems such as a weak lip seal, an uncoordinated suck swallow-breathe pattern, problems with positioning for feeding and severe fatigue were observed, all of which were mainly associated with reduced muscle tone. The potential consequences of these difficulties lead to problems such as coughing, choking, aspiration, vomiting and weight loss.

Newman (2006) also reported that breast feeding is difficult in infants and toddlers with Down syndrome due to the marked hypotonicity, small mouth size, less demanding personality, cardiac defects, and intestinal problems. Later Thomas, Marinelli, and Hennessy in 2007 added that hypotonia can make breast feeding difficult since it impacts sucking behavior. Along with several other disorders that impair neurologic function, Down syndrome can also result in a poorly sustained and depressed sucking reflex.

Schieve, Boulet, Boyle, Rasmussen, and Schendel in 2009 reported that children with Down syndrome are more prone to become sick (head/chest colds or stomach/intestinal illnesses), visit a hospital, go to the emergency room, and require surgery or other medical procedures and according to them, any of these anomalies or

health issues could interfere with breastfeeding. Sooben in 2012 reviewed seven studies from seven different countries spanning over 30 years in order to examine the factors that influenced breastfeeding among mothers of infants with Down syndrome. The review indicated that these children with Down syndrome presented with a range of difficulties in breast feeding during the first few days of life, as a consequence of anatomical, craniofacial, structural abnormalities and medical issues. A delay in introducing solid food to children with Down syndrome was also observed.

Feeding problems in infants with Down syndrome change as they grow older. The infants may experience problems with transition from breast/bottle feeding to cup feeding, and from liquids to solids (Kumin, 1994; Pipes, 1995; Cooper-Brown et al.,2008). Mohamed, Alhamdan, and Samarkandy (2013) found difficulties in using utensils, chewing and swallowing difficulties, food rejection and refusal in children with Down syndrome. They also found that during infancy period, nearly half of the breast-fed Down syndrome children were fed for a duration of less than 6 months, besides 36.4% of the Down syndrome children were bottle fed compared to only 5.5% of the normal siblings indicating the difficulties these children had in transition from bottle feeding to cup feeding, and from liquids to solids.

Chewing was found to be affected in a clinical evaluation of eating behavior conducted by Gisel, Lange, and Niman (1984) wherein they video recorded 26 children with Down syndrome when eating a meal. They found that in children with Down syndrome, chewing behavior was characterized by forward placement of the tongue in the mouth, absence of the maturational changes that occurred in the control participants, and prolonged duration of each masticatory cycle.

A study by Spender Stein, Dennis, Reilly, Percy, and Cave in 1996 found that children with Down syndrome had significantly greater oral-motor dysfunction and an increased tendency to have difficulty with solid foods than a comparison group of children without Down syndrome. They also displayed a delayed acceptance of food, poorly coordinated movement of food from lips into pharynx on eating pureed or solid textures, delayed initiation of feeding sequences for solid and cracker texture, and an overall decreased control of the jaw. Together they represented the aspects of impaired muscle coordination unique to children with Down syndrome. Other studies have also revealed that the acquisition of oral-motor coordination necessary for normal feeding is delayed in children with Down syndrome (Pipes & Holm 1980; Cullen et al., 1981).

Abnormalities in both oral and pharyngeal phase of swallow have been reported. A retrospective study was conducted by Frazier and Friedman (1996) in order to describe the swallowing functions in children with Down syndrome and to identify the possible factors leading to respiratory health issues in these children. The swallowing behavior of 19 children with Down syndrome suggested that the oral phase in these children may be impacted by oral hypersensitivity which can interfere with their acceptance of textured foods. A disordered pharyngeal phase was identified in 16 of the children, with aspiration occurring in 10 of the 19 children studied. Aspiration identified was silent for eight of these 10 children with cough data, and did not correlate with the severity of their oral phase. The study also revealed the difficulties faced by these children with advancing textures, reduced acceptance of food tastes, temperatures and smells which could be attributed to poor control of tongue movement which resulted in gagging and rejection of

age appropriate textured food. These children also faced difficulties in swallowing solids due to the small oral cavity, laxity of the supportive musculature and enlarged tonsils.

Kumin and Bahr in 1999 studied the patterns of feeding, eating and drinking in young children with Down syndrome with oral motor concerns. The study revealed different degrees of hypotonia in different oral structures such as lips, tongue, jaw and found loose ligaments in the temporomandibular joint. The tongue was protruded during spoon feeding and on foods that required chewing. These children also demonstrated limited tongue retraction during swallowing. The authors also found that young children with Down syndrome faced problems in sensory awareness and feedback which impacted feeding, eating, drinking and speech production.

Hennequin, Allison, and Veyrune in 2000 reported that the development of suckling, swallowing, and chewing was delayed and remain impaired in children with Down syndrome. The feeding pattern was found to be any intermediate situation between a primary suckle–swallow pattern and full rotary chewing. Ineffective and depressed control of food between the jaws, lips, and tongue was also observed. They concluded that this group had a high level of functional and dysfunctional oral health problems some of which did not even improve with age.

Field, Garland and Williams in 2003 discovered a significantly higher prevalence of oral-motor problems, swallowing difficulties, and texture selectivity in the 26 children with Down syndrome. They suggested that children with Down syndrome had the ability to chew but refused to do so as a result of learned aversions to specific textures that prompted unpleasant experiences such as gagging and vomiting.

Abnormalities in esophageal phase have also been reported by several researchers. Craig, Peter, and Joyce (1982) found in seven children with Down syndrome abnormalities in esophageal function. Three had recurrent episodes of pneumonia from gastroesophageal reflux; two of these and one other patient had esophageal strictures. Two patients with Down syndrome showed significant abnormalities in esophageal peristalsis with no evidence of gastroesophageal reflux. Literature reports a high incidence of oesophageal atresia, and tracheoesophageal fistula in children with Down syndrome. In 2002, Bianca, Bianca, and Ettore confirmed 90 cases of oesophageal atresia (0.03%) from 1991 to 1998 in 243,916 live births. Other studies also revealed esophageal motor disorders such as abnormalities in esophageal peristalsis and in lower esophageal sphincter function leading to loss of food, food refusal, vomiting, gastroesophageal reflux disease, choking and weight loss (Wallace, 2007).

Need for the study

A look into the literature indicates that feeding problems are quite common in infants and children with Down syndrome. They therefore need to be identified at the earliest so that treatment can be initiated at the earliest to reduce the risk of further complications. If left unattended, feeding disorders in children with Down syndrome may have important long-term health consequences, including growth deficits and decreased performance on academic and cognitive tests. Some feeding problem may persist and can then become habitual if it is not corrected at a very early stage of the child's life, which could lead to the development of behavioral issues that can not only be difficult for the individual as well as produce problems in social integration. Further these could have a negative impact on the quality of life of the child and on his/her parents/caregivers as

well. The in depth assessment of feeding skills will provide valuable input to the speech-language clinician during the treatment of feeding problems in children with Down syndrome. The clinician will be aware of the extent of child's feeding problems in greater detail. This would help the speech-language clinician in planning and prioritizing the goals during therapy. The information will also help in counseling the caregivers, deciding the success or failure of feeding therapy and thereby help in predicting the prognosis of the child.

Although some western studies have been carried out to identify the nature of feeding problems in children with Down syndrome, these are limited. Most of these studies are on infants and focus on the breastfeeding difficulties and oromotor deficits in them. A very few studies objectively assess feeding difficulties in the oral and pharyngeal phases. There are much lesser studies which objectively assess all the three phases of swallow to assess feeding related issues. Hence this study would add to the objective evaluation of feeding difficulties in all the three phases of swallow in children with Down syndrome. Further, as mentioned above, most of the studies have been carried out on infants with Down syndrome. Since feeding is a skill that develops by 2 years of age and refines till 6 years of age (Delaney & Arvedson, 2008), it is essential to study the children in this age group as well. However such studies in the Indian context are limited. The paucity of literature makes it clear that there are deeper underlying complex issues pertaining to feeding in children with Down syndrome that needs to be investigated. A more in depth study covering various aspects related to feeding is to be assessed. No study has undertaken the in-depth quantitative assessment of all the three phases of swallowing. A more effective and practical assessment of a child's ability to feed would

account for the efficient management and reasonable adjustments to be made. Hence the present study was planned to explore in detail the nature of the feeding patterns in children with Down syndrome.

Aim of the study

The present study aimed at assessing feeding problems, if any, in children with Down syndrome in the age group of 2-7 yrs. The specific objectives of the study were

- To develop a questionnaire to assess the feeding problems in children with Down syndrome.
- To investigate the feeding problems, if any, in children with Down syndrome and to compare it with the feeding abilities in chronologically age matched typically developing children.
- To compare feeding abilities between younger and the older group of children with Down syndrome and typically developing children
- To compare feeding abilities across gender in children with Down syndrome and typically developing children.
- To investigate the effects of intervention on feeding abilities in children with Down syndrome.

Chapter 2

Review of Literature

Feeding is of prime importance within the living experience of the young infant and it continues to be a major binding element of experience in the lives of older infants and children. Feeding, swallowing, and respiration are fundamental activities that are essential for the survival, growth, development, nutrition and overall well-being of neonates and young infants. These activities which intersect in the upper aerodigestive tract, represent the most complex neuromuscular unit in the human body. Feeding and swallowing represent the first “window” in which parents and caregivers can view and assess the overall health and neurodevelopmental well-being of neonates and young infants.

Feeding encompasses the process of obtaining food, ingesting the food into the mouth, and swallowing. In normal infants, feeding skills evolve in a sequence that parallels the development of other motor skills and becomes increasingly independent with advancing age. Oral-motor development which supports feeding advances in a stepwise manner starting with the suckle reflex to move on to acquire the more complex oral-motor milestones of sucking, munching, and chewing (Ogg, 1975; Bosma, 1986). The emergence of each oral-motor milestone is dependent upon maturation as well as on successful practice (Illingworth & Lister, 1964; Pinningson & Hegarty, 2000; Eicher, 2002). By four months of age the biologically driven suckle reflex starts to fade away (Ingram, 1962). Infants master suckling and its coordination with breathing when they use the suckle reflex successfully with breast or bottle feeding (Herbst, 1983). This

practice occurs simultaneously along with the neuronal growth and development that together enable the infant to acquire voluntary control over the suckle response (Bosma, 1986; Miller, 1993; Arvedson & Lefton-Greif, 1996).

The dynamic influence between the child's practice during feeding and oral-motor skill development continues until the child has accomplished the most advanced skill of rotary chewing (Smith, Weber, Newton, & Denny, 1991). Increasingly complex tongue movements and oral-motor skill development is integrally linked (Gisel, Schwartz, Petryk, Clarke, & Haberfellner, 2000; Morris & Klein, 2000). In the typical pattern of development, the tongue first moves liquids through a nipple in an anterior/posterior (in/out) pattern (i.e., suckling); and then liquids and pureed foods with a superior/inferior (up/down) pattern (i.e., sucking; Tamura, Matsushita, Shinoda, & Yoshida, 1998; Ayano, Tamuro, Ohtsuka, & Mukai, 2000). The tongue moves chewable foods with a lateral (side to side) pattern over to the molar surface and back to the center, splitting and separating the food until it is ground down enough and recollected to swallow (Ayano et al., 2000). As the child's oral motor function advances, s/he learns to stabilize the jaw, working the tongue off this stable base first centrally with sucking and then laterally with munching (Meyer, 2000; Morris & Klein, 2000). The sweeping action of the tongue, anteriorly, posteriorly, laterally, as well as elevation is achieved with an increase in the range of tongue movements.

The jaw movements, during the initial stage of chewing development, between 6 and 9 months consists primarily of vertical jaw movement along with the suckling motion by the tongue (Gisel, 1991). The complexity of jaw movements increases as the lateral movements of the tongue increases, which helps to transfer the bolus to the molar or

chewing surfaces. On introducing different textured food the overall range of jaw movement also increases (Gisel, 1991). With increase in age and advancement of texture the overall efficiency of chewing improves. With increase in gain of chewing efficiency, children tend to use fewer chewing cycles, along with a decrement in chewing duration across the transition period. It is only after 3 years of age the chewing duration stabilizes (Gisel, 1988). Studies have revealed that by 6 to 24 months of age children are able to maintain circumoral (lip) movements during feeding and use full lip closure on a spoon for food removal as well as to retain a bolus in the oral cavity. In children the achievement of lip closure to remove a bolus from the spoon or to retain a bolus within the oral cavity varies by texture and age. All children by 12 months of age achieve lip closure for all consistencies for both removal and retention. In summary, the child's feeding experience and practice directly influence the oral-motor pattern and oral-motor pattern directly influences feeding response (Bosma, 1986).

Phases of Normal Feeding

Oral feeding and swallowing depend on a highly complex and integrated sensorimotor system that involves three distinct anatomic regions, including the oral cavity, pharynx, and esophagus. Feeding and swallowing process for liquids and food can be functionally divided into four phases.

Oral Preparatory Phase

Sensory recognition of food approaching the mouth and being placed in the mouth is critical before any oral preparatory movements can be initiated. The oral preparatory phase is voluntary and its length varies considerably, depending on the viscosity of the material to be swallowed and the amount of oral manipulation the individual uses in

savoring a particular food. This phase is further divided into two phases i.e. transfer phase and reduction phase. During the transfer phase, the tongue arranges the bolus and moves it posterior where it can be chewed. In normal individuals, transfer phase usually results in the food being placed in region of the molar teeth. Following this, the reduction phase takes over which involves the manipulation of food in the mouth to form bolus. This phase requires the mastication of materials, which involves rotary lateral movements of the mandible and tongue. The tongue positions the materials on the teeth. When the upper and lower teeth have met and crushed the material, the food falls medially towards the tongue, which moves the material back onto the teeth as the mandible opens. The cycle is repeated numerous times before forming a bolus and initiating the oral phase of swallow. In addition to this cyclic movement during mastication, the tongue mixes the food with the saliva (Lowe, 1980).

In infants, sucking and swallowing of liquids are accomplished with minimal time in this phase. As children begin to handle thicker, chunkier textures, the oral preparatory phase may last for several seconds. The more chewing that is required, the longer this phase lasts. Oral manipulation of liquid presented via cup varies significantly from one child to another, but usually liquid is held in the oral cavity for no more than 2 to 3 seconds. Lip closure is needed once the material is placed into the mouth so that no liquid will dribble down the chin. Some children may move the liquid around in the mouth before they form a cohesive bolus. The bolus is then held between the tongue and hard palate before the initiation of voluntary swallow. During this phase, the soft palate is in a lowered position, helping to prevent the bolus of liquid from entering the pharynx before the swallow is produced. This active lowering of the soft palate occurs by contraction of

the palatoglossus muscle. The larynx and pharynx are at rest. The airway is open and nasal breathing continues until a swallow is produced.

Oral Phase

The oral phase is a voluntary phase, which begins with the posterior propulsion of the food bolus by the tongue and ends with the production of a swallow. The initiation of a swallow of a bolus of food or liquid is under voluntary control, although the final stages of the swallow process are involuntary. The voluntary actions in manipulating a bolus of food or liquid include the elevation of the tongue followed by a posteriorly directed movement resulting in a peristaltic motion. The tongue movement during this oral phase has often been described as a stripping action, with the midline of the tongue sequentially squeezing the bolus posteriorly against the hard palate. The sides and tip of the tongue remain firmly anchored against the alveolar ridge. During this time, a central groove is formed in the tongue, acting as a ramp or chute for food to pass through as it moves posteriorly (Ramsey, Watson, Gramiak, & Weinberg, 1955). As food viscosity thickens, the pressure of the oral tongue against the palate increases, requiring greater muscle activity (Dantas & Dodds, 1989). Thicker foods require more pressure to propel them cleanly and efficiently through the oral cavity and pharynx (Reimers-Neils, Logemann, & Larson, 1994). Elevation of the soft palate against the posterior pharyngeal wall occurs as the bolus enters into the pharynx. The bolus leaves the mouth and at the same time the nasopharynx is sealed off to prevent nasopharyngeal reflux. The oral phase timing does not vary according to texture and lasts less than 1 second in normal individuals.

Pharyngeal Phase

The pharyngeal phase of swallowing is involuntary and begins with the production of a swallow and the elevation of soft palate to close off the nasopharynx. The pharyngeal phase consists of peristaltic contraction of the pharyngeal constrictors to propel the bolus through the pharynx. Simultaneously, the larynx is closed to protect the airway. The larynx protects the airway in two major ways. The most important protection is the complete and automatic closure of the glottis during swallowing. The epiglottis is brought down over the glottis during swallowing and deflects the bolus of swallowed material laterally and posteriorly towards the upper esophageal segment. The second major protective function of the larynx is the production of the protective cough reflex which is triggered by sensitive receptors stimulated by the vagus nerve in both the larynx and subglottic space. Shaker, Dodds, Dantas, Hogan, and Arndorfer (1990) have noted four sequential actions related to laryngeal closure i.e.

- i. Adduction of the true vocal folds associated with the horizontal approximation of the arytenoid cartilage.
- ii. Vertical approximation of the arytenoids to the base of the epiglottis.
- iii. Laryngeal elevation, and
- iv. Epiglottis descent.

During swallowing as the epiglottis moves downwards, sphincter contraction of intrinsic laryngeal muscles occurs to approximate the arytenoids and epiglottis and to close the false and true vocal folds. At the same time the larynx is elevated and pulled

forward, away from the bolus path. The bolus is propelled to the esophagus by contraction of the pharyngeal muscles. During pharyngeal contraction, the larynx elevates, the glottis closes, and respiration ceases to protect the lower airway from aspiration. Because the pharynx is the common chamber for the respiration and digestive pathways, important developmental changes affect the ability to swallow safely. A peristaltic wave of contraction of the pharynx propels the bolus into the esophagus. During passage of the bolus through the pharynx, excellent coordination between breathing and swallowing is essential to prevent aspiration.

Esophageal Phase

The esophageal phase consists of an automatic peristaltic wave, which carries the bolus to the stomach, thus reducing the risk for gastroesophageal reflux, or reentry of material from esophagus into the pharynx. Gastroesophageal reflux is also prevented by the tonic contraction of the cricopharyngeus muscle. The process of peristalsis moves the bolus through the esophagus and ends when the food passes through the gastroesophageal junction. At birth, the greater pressure in the esophagus is the principal mechanism of preventing reflux of the stomach contents (Boix-Ochoa & Canals, 1976). In the first few weeks after a term birth, these anatomic mechanisms of closure at the gastroesophageal junction mature rapidly. Thereafter, the pattern of esophageal swallow peristalsis is essentially the same in infants, children and adults. An esophageal phase promptly follows each separate pharyngeal phase of swallow when there is definite time delay between swallows (Ingelfinger, 1958; Dodds, Hogan, Reid, Stewart, & Arndorfer, 1973). Studies have found that swallow induced peristalsis normally propagates at about 2 to

4cm/sec and transverse the entire body of the esophagus in 6 to 10 sec (Arvedson & Brodsky, 1993).

Development of Feeding

From birth on, a child adapts to a changing environment. Through brain development and learning experiences, children begin to take control of their world. Motor development is the primary basis on which children learn to adapt, interact and manage their environment. There is a natural order of motoric events that are precursors to the stages of movement; similarly feeding progression also follows a similar order of events that are intricately connected to motoric development. The development of feeding and swallowing is the result of a complex interface between the developing nervous system, various physiological systems and the environmental factors that begin in embryologic and foetal periods and continue through infancy and early childhood. The development of feeding is sustained by neurological maturation and ongoing experiences of a child. Most children negotiate the necessary developmental sequence without significant difficulties. Acquisition of age-appropriate feeding skills is central to the infant's development of self-regulation and independence.

Feeding is a learned skill, where the development of suck swallow response in the newborn to the adult pattern of chewing and swallowing is not prewired. Infants require the appropriate exposure to the increasing challenges produced in handling more difficult types of food. Stevenson and Allaire (1991) summarized the following statements as the major elements involved in the development of feeding and swallowing

- i. Structural integrity is essential to the development of normal feeding and swallowing skills. The anatomic changes associated with growth and development affect feeding function in children.
- ii. Normal infant feeding process is reflexive in nature which is under brainstem control, and does not require suprabulbar input. As feeding development progresses, basic brainstem-mediated responses come under voluntary control.
- iii. The mature swallow consists of a voluntary oral-preparatory phase, a voluntary oral phase, and involuntary pharyngeal and esophageal phases whereas the infant swallow does not have a voluntary oral-preparatory and oral phase but is otherwise similar.
- iv. The neurophysiologic control of feeding and swallowing is complex and involves sensory afferent nerve fibers, motor efferent fibers, paired brainstem swallowing centers, and suprabulbar neural input. Close integration of sensory and motor functions is essential for the development of normal feeding skills.
- v. Feeding development, although dependent on structural integrity and neurologic maturation, is a learned progression of behaviors. This learning is heavily influenced by oral sensation, fine and gross motor development, and experiential opportunities.
- vi. The basic physiologic complexity of feeding is compounded by individual temperament, interpersonal relationships, environmental influences and culture.

- vii. The main goal of feeding is the acquisition of sufficient nutrients for optimal growth and development of the child.

The major systems that must be present and functional for normal feeding are summarized in the table 2.1 given below.

Table 2.1

Systems and domains involved in the development of feeding. (Source: Arvedson & Brodsky, 1993; Stevenson & Allaire, 1996)

Systems	Required for
Oromotor function	Sucking, munching, chewing, and movement of the bolus; also needed for speech
Respiratory system	Maintaining normal oxygen exchange, coordinating suck and swallow, coughing to protect airways
Cardiovascular system	Maintaining normal blood pressure and oxygenation of the tissues
Pharyngeal coordination	Coordinating swallowing and breathing, safely transporting the bolus to the esophagus
Gastrointestinal system	Esophageal transporting of the bolus to the stomach and lowering esophageal sphincter to avoid reflux. Gastric emptying to the duodenum and transporting throughout

	the bowel
Gross motor domain	Maintaining head in midline and upright position, sitting stability on the chair
Fine motor domain	Finger feeding, using a spoon, holding a cup
Expressive language domain	Asking for more or saying no during feeding
Nonverbal Communication games	Pointing for food, opening mouth to receive food, gesturing, playing
Receptive language domain	Comprehension of the meaning of words, “food, bottle”, understanding of commands
Hypothalamus	Controlling hunger and satiety
Cognitive domain	Recognizing foods by color, appearance, taste and so on; learning the associations related to feeding; learning to self-serve food
Social domain	Giving positive feedback to the caregiver, eye contact
Caregiver (socioeconomics)	Providing appropriate amount and type of food
Caregiver (emotional)	Funneling positive emotional support of a child during the learning process, setting rules and limits

As seen from the table, feeding development is related to the development of the oral structures, upper limbs and other skills which determines the pattern of feeding. New born infants with intact anatomy and neurologic function most often quickly become efficient feeders. Compared to an older child, infants are only able to suck and swallow liquids and these preliminary feeding skills are very essential to their existence. The oral and pharyngeal cavities are smaller because of the relatively small mandibles and fat pads in the cheeks; the tongue fills the oral cavity and seems specifically designed to hold a nipple in place for feeding. As the child grows, the sucking and swallowing action is followed by biting, chewing, eating from a spoon, drinking from a cup and a straw which are more mature feeding behaviors. These developments occur as the higher cortical centers gain more control (Arvedson & Brodsky, 1993). The normal progression seen in the development of feeding has been described below:

Suckling and Sucking

Two distinct phases of suck occur in infant development, suckling and sucking. Suckling, the earliest intake pattern observed in infants is gradually acquired in the 2nd and the 3rd trimesters of gestation. The suckle action, involves the coordination of the tongue, hyoid, mandibular muscles and the lower lip. Suckling involves the definite backward and forward movement of the tongue such that suction and nipple compression succeed each other (Morris & Klein, 1987). Liquid is drawn into the mouth through a rhythmical licking action of the tongue, combined with pronounced opening and closing of the jaw. Lips may be closely approximated. The tongue moves forward for half of the suckle pattern, but the backward phase is the most pronounced. Tongue protrusion does not extend beyond the border of the lips. Sucking from the breast of the mother is what

the infants learn to do first. During breast feeding, the tip of the tongue stays behind the lower lip and over the lower gum, while rest of the tongue cups around the areola of the breast. The mandible moves the tongue up, allowing the breast areola to be compressed against the infant's alveolar ridge. Milk is then expressed into the oral cavity (Arvedson & Brodsky, 1993).

Sucking is the second intake pattern to develop between 6 and 9 months. In this type of feeding pattern, the tongue body raises and lowers with strong movement of its intrinsic muscles, and thus jaw makes a smaller vertical excursion (Morris & Klein, 1987). The developmental sequence of suckling and sucking is one of the steps in preparation for oral manipulation of thicker liquids and soft food with spoon feeding.

Transitional Feeding

The transitional feeding period is a time of progressive gradation from exclusive suckles feeding of liquids to voluntary ingestion of physically varied food materials. The transitional feeding phase typically starts at 4-6 months of age in normal infants. The inclination for different textures after several months of suckle feeding is primarily related to changes in the central nervous system along with some anatomical changes. The increasing variety of taste and smell of the foods offered to the infants may be one of the major factors in the achievement of transitional feeding (Bosma, 1986). There is an increase in intraoral space as the mandible grows downward and forward. The oral cavity also elongates in the vertical dimensions. The hyoid bone and larynx shift downward, so that the breathing and swallowing coordination becomes a factor during feeding, and breathing and swallowing truly become mutual activities (Arvedson & Brodsky, 1993).

Eruption of teeth maybe the most notable change in the peripheral anatomic structures. Deciduous teeth erupt between 6 to 24 months after birth, with all 20 deciduous teeth usually present by the end of the second year in healthy children. Mandibular incisors usually erupt 6-8 months after birth but the process may be as late as 12 to 13 months in some normal children (Moore, 1988). Molars erupt from 12 to 24 months and the canines from 16 to 20 months. The erupted teeth are probably more important as sensory receptors than for motor purposes, because biting and chewing during transitional period can be accomplished effectively with no teeth. Bosma (1986) has suggested that the sensory input of the teeth may be sufficient in the development of central nervous system control of the feeding process. In this phase the buccal cavity space increases, food is manipulated between the tongue and the buccal wall. It is especially common for infants to move toward the first spoon experiences with suckling movement of the tongue. Sometimes, this movement may appear to be tongue thrusting. Progressively the lateral tongue action becomes sturdier along with the rotary jaw action required for efficient oral stage.

The introduction of spoon feeding of thin smooth pureed food in infants occurs around the age of 6 months. After all single ingredient foods have been offered, by 7 and 9 months of age combination of foods that are smooth pureed foods is given. Gradually food with texture such as dissolvable solids (e.g., soft biscuits at 6-9 months), textured puree foods (e.g., mashed banana at 6-9 months), ground solids at 6-9 months, soft diced solids (e.g., fruits and vegetables at 9-12 months) is introduced. Eventually a general toddler diet of table foods is given by 12-18 months of age (Delaney & Arvedson, 2008). During spoon feeding early transition feeders are described as using wide and ungraded

jaw movements. Due to lack of experience, while accepting the bolus the opening movements of the jaw are inaccurate and overshoot the intended target. As a beginning step, cup drinking is usually introduced by the age of 6 and 9 months, in order to wean from breast and bottle feeding. By the age of 12 months children generally receive their fluids through a combination of bottle or breast feeding and cup with a valve or a straw. By their first birthday children are expected to drink independently from a spout cup or straw. Independently drinking from an open cup usually occurs much later.

The initial stages of chewing development which is established between 6-9 months consists primarily of vertical jaw movements (munching) along with a suckling motion of the tongue on chewing solids. With the lateral movements of the tongue being established, the complexity of jaw movements increases, simultaneously transferring the bolus to the molar or chewing surfaces. The advances of texture also increase the range of jaw movements. With increasing age and introduction of varied texture of foods, the children gain chewing efficiency and use fewer chewing cycles. Chewing duration for solids decreases significantly across the transition periods and stabilizes only after 3 years of age. By 12 months of age all children achieve lip closure in order to remove bolus from the spoon to retain a bolus within the oral cavity. The tongue continues to be a primary structure for oral feeding. As infant mature towards semi-firm food, the tongue moves the food to the lateral buccal area where it is mashed by vertical motion of the tongue and jaw. These manipulations probably are a prelude to molar chewing. Children gradually wear out of the suckling pattern from 6-10 months of age for viscous consistency as well as from 6 to 12 months of age for puree. The motions of chewing occur with or without erupted molars in young children.

In the course of development, vertical movements happen to be related with alternating lateral motor features of mature mastication. The children start making distinct lateral shifting of bolus from midline on to the molar surfaces and again back to the midline (Morris & Klien, 2000). From 6-24 months of age, children lateralize the bolus from one molar surface to the other in smooth and coordinated movements, for solids and not for food of viscous consistency (Stolovitz & Gisel, 1991; Morris & Klien, 2000). However only a small percentage of children over 2-5 years precisely move solid food from side to side, instead they tend to have a preference on slow and rolling tongue movements to lateralize food (Gisel, 1988). Mastication coordination becomes fully mature between 3-6 years of age (Vitti & Basamajian, 1975). From 12-36 months of age children refine their oral skills, expand the different texture of foods they accept, become more precise and efficient at chewing food that require more extensive oral manipulation and also start handling liquids from open cups (Delaney & Arvedson, 2008). The feeding progression from birth to 24 months has been illustrated in the table 2.2 below.

Table 2.2

Feeding progression from birth to 24 months. (Source: Cichero & Murdoch: Dysphagia: foundation, theory and practice, 2006)

Age	Food	Oral Preparation and Oral Events	Feeding Utensils
Birth to 6 months	Milk, Liquids	Suckling and then sucking	Breast or bottle
4-6 months	Cereals, puree	Initially sucking, the tongue to palate movement; may eject food from spoon involuntarily; gags on new textures	Spoon
6-9 months	Chunky puree, mashed food, soft finger foods	Emerging munching pattern, desensitization of gag reflex; lateralization of food to gums; deciduous teeth erupting	Spoon; drinking from cup (at 9 months)
9-12 months	Chopped food and finger food	Licking food off lips; biting of objects; controlled, sustained bite on hard food (e.g., biscuit/	Spoon, cup; self feeds with fingers; weaning from breast/ bottle as cup

		cracker)	drinking increases
15-24 months	Full diet with some exclusionary items (e.g. Nuts)	Licks food from lips, increased maturity of adult rotary chew pattern, and jaw stability in cup drinking; independence in self-feeding; straw drinking.	Spoon, cup, fork; self-feeding predominates.

Neurodevelopmental Milestones Relevant to Normal Feeding

Maturation of the central nervous system (CNS) plays an important role in the acquisition of normal swallowing or feeding skills. Both feeding and swallowing require intact functioning of the central and peripheral nervous systems and the intricate coordination of actions of multiple muscles of the oral cavity, pharynx and esophagus (Miller, 1982). As the brain develops throughout the first several months of life, sensory inputs pertinent to feeding extend into the midbrain, cerebellum and thalamus and to the central cortex (Arvedson & Brodsky, 1993). The sense of taste and smell also has a significant role in feeding. A child's neurodevelopmental status determines the development of feeding and swallowing.

The development of functional feeding skills reflect the tremendous achievements in sensorimotor integration of swallowing and respiration, hand-eye coordination, normal muscle tone and posture, and appropriate psychosocial maturation during the first three

years of life (Arvedson & Brodsky, 2002; Carruth & Skinner, 2002). Feeding development does not occur in isolation. There are many other skills such as cognitive and sensory motor skills that develop simultaneously in the child which would in turn facilitate feeding skills. Many aspects of feeding are manifestations of normal cognitive and motor development. The neurodevelopmental milestones relevant to normal feeding has been summarized in the table 2.3.

Table 2.3

Neurodevelopmental milestones relevant to normal feeding. (Source:Arvedson& Brodsky, 1993)

Age (months)	Cognitive skill	Sensory- Motor Skill	Feeding Skill
Birth -2	Visual fixation and tracking	Balanced flexor and extensor tone of neck and trunk	Promotion of parent-infant interaction during feeding
3-4	Visual recognition of parents	Head maintained primarily in midline and aligned with trunk in supported sitting.	Parents preferred for oral feeding Upright supported posture for spoon feeding
5-9	Visual interest in	Independent sitting	Feedings more

	small objects	Extended reach with	frequently in
	Object permanence	pincer grasp	upright posture.
	Stranger anxiety		Initiation of finger feeding
			Parents preferred for feedings
18-24	Use of tools	Refinement of upper extremity coordination	Use of feeding utensils
	Increasing attention and persistence in play activities		Prefer to feed self over longer period of times
	Independence from parents		Imitate others during meals
	Parallel or imitative play		

Factors Influencing the Development of Feeding

Feeding and swallowing movements and behaviors are very complex and clearly involve much more than just activities in the mouth, throat and stomach (Gisel, Birnbaum, & Schwartz, 1997). The sensory motor systems provide both the structural foundation and the sensory information that enable a child to practice and master oral-motor skills (Morris & Klein, 2000). Structural alignment, control and sensory input are

important factors in influencing development of feeding as the dynamic feeding process involves internal activities such as breathing, digestion and elimination.

Oral feeding skill may be different in different individuals. It is influenced by the levels of alertness, stress, illness and fatigue as well as actual motor coordination in the oral pharyngeal system. Also, the cultural patterns along with social factors within the family have an effect on the feeding skills. The transitional feeding time period is the phase at which, the dietary pattern and preferences are acquired, and many of these preferences are carried over into adulthood (Arvedson & Brodsky, 1993). The aspect of anatomy, embryology and physiology a human embraces also influences the feeding development. In addition, feeding is influenced by behavioral and social factors (Miller, 1993).

Feeding Problems in Infants and Children

Feeding disorders affect children with normal development as well as those with developmental disabilities, which in turn can lead to malnutrition and respiratory symptoms. Any abnormalities of developing brain or structural or functional deficits in the oral, pharyngeal or esophageal region commonly results in a spectrum of cognitive, communicative, behavioral and motor abnormalities that are often associated with feeding and swallowing disorders. Even a small injury to the developing brain can have a magnified negative effect on the rest of the sequence of the developing brain leading to congenital malformation and other related abnormalities. About 25% of children are reported to present with some form of feeding disorder, which increases to 80% in developmentally delayed children. Estimates suggest as many as half of parents of

toddlers report feeding issues at some time during the early years, with children with special needs (medical, developmental, and physical) being even more at risk for developing feeding problems that require intervention. General population surveys using parental interviews have revealed an incidence of minor feeding problems ranging between 25% and 35% in normal children and with more severe feeding problems observed in 40% to 70% of infants born prematurely (Hawdon, Beauregard, Slattery, & Kennedy, 2000) or children with chronic medical conditions. Dahl (1987) reported that of some of these difficulties were transient in nature, however some problems such as refusal to eat, found in 3% to 10% of children tend to persist into adulthood.

Causes of Feeding Problems

Feeding problems can result from the adverse effects of various medical conditions, congenital problems or developmental issues. Most feeding disorders have underlying organic causes. However, many studies indicate that underlying cause of abnormal feeding patterns are not mainly due to an organic impairment. Hence feeding disorders can be conceptualized on a continuum between psycho-social and organic factors. Burklow, Phelps, Schultz, McConnell, and Rudolph (1998) classified complex pediatric disorders based on the Interdisciplinary Feeding Team (IFT) at Children's Hospital Medical Center in Cincinnati, Ohio by a team of Pediatric Gastroenterology, Nursing, Nutrition, Occupational Therapy, Psychology, and Speech Pathology. They developed a classification system based on a large sample of children who were presented for an evaluation of complex feeding problems. The following five categories were identified by the IFT as reflecting the nature of the complex pediatric feeding disorders.

- i. **Structural Abnormalities:** Anatomic abnormalities of the structures associated with eating and feeding, for e.g., defects associated with Pierre-Robin syndrome such as retrognathic jaw, cleft palate and posterior tongue placement, macroglossia, tracheotomy, esophageal strictures or stenosis.
- ii. **Neurological Conditions:** Feeding problems associated with CNS insult or musculoskeletal disorders, for e.g., cerebral palsy, muscular dystrophies, cranial nerve dysfunction, mental retardation/developmental disabilities, brain stem injury, pervasive developmental disorder.
- iii. **Behavioral Issues:** Feeding difficulties resulting from psychosocial difficulties (poor environmental stimulation, dysfunctional feeder-child interaction), negative feeding behaviors shaped and maintained by internal and/or external reinforcement (selective food refusal, rumination), and/or emotionally based difficulties (phobias, conditioned emotional reactions, depression).
- iv. **Cardiorespiratory Problems:** Feeding difficulties associated with diseases and symptoms which compromise the cardiovascular and respiratory systems, complicating the coordination of sucking, swallowing and breathing during feeding , for e.g., tachypnea associated with bronchopulmonary dysplasia.
- v. **Metabolic Dysfunction:** Feeding difficulties associated with metabolic diseases and syndromes which interfere with the development and/or maintenance of normal feeding patterns, for e.g., hereditary fructose intolerance, Dumping syndrome, Down syndrome.

Children with genetic syndromes such as CHARGE syndrome, Noonan syndrome, Pierre Robin syndrome, Velocardiofacial syndrome, Williams syndrome, Di

George syndrome, Cornelia de Lange syndrome, Klippel Feil, Sotos syndrome, Prader Willi syndrome and Down syndrome frequently have feeding problems and swallowing dysfunction (Samuels & Chadwick, 2006). These problems arise as a result of the complex interactions between medical, anatomical, physiological, and behavioral factors. Feeding problems which are associated with genetic conditions may cause feeding to be an unpleasant or negative experience or even painful because of choking, coughing, gagging, fatigue, or emesis. This can result in the child to stop eating and to develop behaviors during feeding that make it difficult, for a parent to feed their child. In addition, the related medical or physical conditions or other variables such as prematurity limits the experiences with oral intake, resulting in a failure or delay in the development of normal oral motor skills.

Amongst these syndromes the feeding difficulties in Down syndrome has been frequently documented in literature. Neuromotor coordination impairments, structural abnormalities such as those associated with Down syndrome (e.g., hypotonia, poor tongue control, and open mouth posture) frequently interfere with the acquisition of effective oral-motor skills and predispose them to potential feeding problems. Along with oral motor problems, frequent dental anomalies such as periodontal disease, tooth loss, severe bruxism, increases the risk of developing feeding problems in them. Several comorbidities like chronic illnesses such as anatomical abnormalities, congenital heart disease, esophageal, stomach, and bowel dysmotility are common problems leading to poor feeding and poor growth in Down syndrome (Cronk, Crocker, Pueschel, Shea, Zackai, Pickens, & Reed, 1981).

Down Syndrome

Down syndrome is the most commonly identified genetic form of mental retardation and the leading cause of specific birth defects and medical conditions. It is usually identified at birth and is confirmed by a karyotype showing trisomy of chromosome 21. Down syndrome is predominantly caused due to non-disjunction of chromosome 21 during meiosis, resulting in a zygote carrying an extra 21st chromosome. Most cases of Down syndrome involve a non-disjunction during the first meiotic cell division, with mothers contributing the extra chromosome in 85% of cases, while translocation of an extra copy of the same chromosome accounts for a small proportion of the condition. Mosaicism comes about when the extra chromosome 21 is present in some, but not in all cells of the individual. Trisomy 21 accounts for 95% of cases translocation for 4% and mosaicism for 1% of all cases. Down syndrome is characterized by central growth deficiency with delayed mental and physical development. All individuals with Down syndrome are mentally impaired to some degree, ranging from mild to severe retardation (Lejeune, 1959).

Epidemiology of Down Syndrome

Down syndrome affects people of all ages, races, and economic levels. Women aged 35 and older have an increased risk of having a child with Down syndrome. Down syndrome occurs once approximately every 800 to 1000 live births. Approximately 5000 children with Down syndrome are born each year and over 350,000 people have Down syndrome in United States (National Down Syndrome Society, 2000). Over the last two decades, Down syndrome epidemiology in Europe and other countries has been influenced by three main factors that have determined a significant modification of

incidence and prevalence, they are: changes in the distribution of maternal reproductive age; the number of terminations of pregnancy after a prenatal diagnosis; and the decreased mortality in the early years of life and then a prolonged survival of persons with Down syndrome. The real incidence of Down syndrome, as well as of other congenital anomalies, is actually unknown since all the conceived zygotes should be evaluated and counted.

Canfield et al., (2006) obtained an estimate of the birth prevalence of Down syndrome in the U.S. The estimated maternal age-adjusted prevalence of Down syndrome based on the surveillance of 22% of the live births in the U.S. was 13.65 per 10,000 live births, or 1/732. This suggested that nearly 5,400 of the 4 million infants born each year in the U.S. have Down syndrome. Sherman, Allen, Bean, and Freeman in 2007 estimated Down syndrome to occur in approximately 1 in 732 infants in the United States. Overall, incidence of Down syndrome worldwide ranges from 1.25–1.67/1000 live birth, making it by far the most common form of chromosomal non-disjunction (Mohamed, 2007). Irving, Basu, Richmond, Burn, and Wren, in 2008 estimated the overall prevalence of Down syndrome in the United Kingdom and found 1.08 per 1000 live births from 1985-2004 and one-year survival of live births with Down syndrome was reported to have increased, especially in babies with cardiovascular malformations, reaching almost 100%. Globally, as of 2010, Down syndrome occurs in about 1 per 1000 births (Weijerman & De Winter, 2010).

Isaac, Krishnamurty, Reddy, and Ahuja (1985) conducted a survey of Down syndrome incidence in Hyderabad and found an incidence of 1.17 per 1000 or 1 in 853 live births. Verma, Anand, Kabra, Menon, and Sharma (1998) found a frequency of

0.81/1000 Down syndrome in Delhi region. A prospective study of 17,653 consecutive births for two years in Mumbai was undertaken to survey an overall incidence of malformations. The incidence of trisomy 21 was 1 in 1200 (Patel & Adhia, 2005).

Clinical Manifestations of Down Syndrome

This genetic condition encompasses a series of many clinical complications such as cardiac, neurological, endocrinal, respiratory, hematologic, ophthalmologic and gastrointestinal problems that affects the overall development of the child (Penrose & Smith, 1966; Miller and Leddy, 1998; Vicari, 2006; Van Gameren-Oosterom, Fekkes, Buitendijk, Mohangoo, Bruil, & Van Wouwe, 2011). Their general physical characteristics include

- i. **Head:** The outstanding feature is brachycephaly with prominence of the forehead, shortening of anteroposterior diameter and flattening of the occiput, the cranial capacity being below than normal.
- ii. **Spine:** Malformations of the spine, particularly the upper cervical region, instability of the atlantoaxial joint, hip dislocations may lead to spinal cord injury. This is due to laxity of the transverse ligaments that hold the odontoid process close to the anterior arch of the atlas.
- iii. **Face:** The round flat face is characterized by a flat nasal bridge, epicanthic folds and palpebral fissures that slant upward. The facial musculature have multiple anomalies such as additional, missing, or poorly differentiated muscles, hyperextendable joints and nerve innervation differences (Miller & Leddy, 1998). These variances seen in muscular innervation are partly thought to account for the reduced speed, limited

range of motion, and difficulty with coordination of the speech articulators observed in individuals with Down syndrome and may impact speech intelligibility.

- iv. ***Nose:*** The nose is small and the nasal bones are underdeveloped and there is a tendency for the nares to point forwards instead of down. Rhinitis is also common (Penrose & Smith, 1966).
- v. ***Eyes:*** The most common findings are hypoplasia of the iris stroma, brushfield spots and epicanthic folds. The palpebral fissures are oblique and narrow laterally. There is persistence of a complete epicanthal fold.
- vi. ***Ears:*** The ears are usually small, low set and simple in appearance, often with an over folded upper helix, poor antihelix development, hypoplastic tragus and a small lobe. The cartilage is often deficient. The diameter of the external auditory meatus is abnormally narrow, which prevents good visualization of the tympanic membrane. Structural anomalies of the middle and inner ear such as permanent fixations of the stapes, congenital malformations of the bones of the middle ear and shortening of the cochlear spiral are seen. Individuals with Down syndrome are susceptible to chronic ear infections and chronic middle ear effusions with associated hearing loss, airway obstruction, and sleep apnea, as well as problems with chronic rhinitis and sinusitis.
- vii. ***Lips:*** Lips have radical furrows as a consequence of the generalized hypotonia.
- viii. ***Mouth:*** Protrusion of the tongue is common in newborns and is attributed to small mouth. The tongue is normal in appearance at birth, but in older children there is hypertrophy of the vallate papillae and a furrowed appearance. The teeth often erupt late and are misshapen and small. Congenital absence of both deciduous and

permanent teeth is common, especially the third molars, second molars and lateral incisors.

- ix. **Neck:** The neck is short and broad and the hairline reaches farther down the back than normal. Infants have looseness of skin over the neck and shoulders, but webbing of the neck is rare. A roll of fat is evident at the nape of the neck.
- x. **Abdomen:** In children the abdomen is protruded and diastasis recti and umbilical hernia are common.
- xi. **Limbs:** The hands, feet and digits are broad and shortened. Clinodactyly of the fifth finger and wide space between the first and second toes are present in majority of the individuals with Down syndrome. The joints in the upper and lower limbs are usually hyperextensive, especially in infants and young children. Occasionally clubfeet is also observed (Vicari, 2006; Van Gameren-Oosterom, Fekkes, Buitendijk, Mohangoo, Bruil, & Van Wouwe, 2011).
- xii. **Dermatoglyphics:** The most common features are a transverse palmar crease(simian crease), a single flexion crease of the fifth finger in association with clinodactyly, a distal axial triradius on the palms, ulnar loops on all fingers, and a tibial arch in the hallucal area of the soles.
- xiii. **Skin:** The hands and feet often have a mottled, and sometimes cyanotic, appearance in infants and adults. The cheeks of infants are usually red and scaly. Generalized rough, dry skin becomes increasingly frequent with advancing age. Xeroderma and localized chronic hyperkeratotic lichenification with reduced Vitamin A levels.

- xiv. ***Hair:*** Beards tends to be slight. Scalp hair is smooth and soft in children and more sparse and rough in older adults. Recurrent focal alopecia is a common problem in adults.
- xv. ***Height:*** The length at birth is below average and height is below the normal range at all ages.
- xvi. ***Heart:*** Malformations affect the heart and its vessels. Defects in anteroventricular septum, patent ductus arteriosus were the most common abnormalities seen.
- xvii. ***Gastrointestine:*** Gastrointestinal tract anomalies such as duodenal atresia or stenosis, anal atresia and megacolon are seen.
- xviii. ***Thyroid:*** Another common abnormality is the dysfunction of the thyroid gland where individuals with Down syndrome tend to present hypothyroidism and it is related to an underdevelopment of the bones and the teeth and to a delayed tooth eruption (Thiel & Fowkes, 2007).

In addition to the above mentioned clinical complications, children with Down syndrome are predisposed for language learning deficits and speech production errors which may be related to differences in oral structure and function (Stoel-Gammon, 1997; Miller & Leddy, 1998). There are also reports of apraxia (difficulty in execution of the motor programming of speech movements) and dysarthria (weakness or incoordination of the articulators that results in slow, weak, imprecise, or disorganized speech) (Miller & Leddy, 1998; Dodd & Thompson, 2001; Kumin & Adams, 2000; Kumin, 2006; Rupela & Manjula, 2007). Speech production errors due to reduced speed, range of motion, and coordination of the articulators leads to poor speech intelligibility. Specifically, syllable

structure phonological processes, such as omission, substitution, cluster reduction and final consonant deletion, appear to be common in children with Down syndrome. The children with Down syndrome demonstrate a delay in acquiring speech and language skills. Research has indicated that speech and language development for children with Down syndrome progresses in an uneven pattern characterized by rapid spurts and changes with long periods of plateau (Miller, 1987). Evidence suggests that receptive language in children with Down syndrome is typically stronger than expressive language. Phonology, expressive vocabulary, receptive and expressive syntax, and some pragmatic aspects of language are impaired.

Cognition is also affected in most children with Down syndrome Carr and Carr in 1995 stated that individuals with Down syndrome have considerable variation in their psychological, and intellectual characteristics, for e.g., the intelligence score in children and adults can vary over 50-60. Nearly 80% of individuals show moderate retardation (Roizen, 2002). Dykens, Hodapp, and Evans (1994) found that Down syndrome exhibit greater lags and impairments in cognitive based communication skills than in interpersonal, social and adaptive behaviors. Cognitive development is usually delayed and learning difficulties persist throughout life. The various qualitative differences in cognitive development include the growing use of avoidance strategies when faced with cognitive challenges, failure to consolidate newly acquired cognitive skills into the repertoire, less than effective use of existing problem-solving skills, an increasing reluctance to take the initiative in learning, difficulty in learning contingencies and greater contentedness with non-contingent reinforcement schedule than typically developing children (Spiker & Hopmann, 1997).

Individuals with Down syndrome also exhibit behavioral and psychiatric problems such as attention deficit/hyperactivity disorder, obsessive-compulsive disorder and depression (Roizen & Patterson, 2003). Earlier studies indicate that nearly 10–12% of individuals with Down syndrome also had a diagnosis of autism spectrum disorders (Ghaziuddin, Tsai, & Ghaziuddin, 1992).

Structural Basis of Feeding Problems in Children with Down Syndrome

Many of the medical and physiological characteristics of Down syndrome have direct consequences on oral health, feeding and swallowing and indirect consequences on the quality of life of persons with Down syndrome and their caregivers. As high as 80% of individuals with Down syndrome are affected by dysphagia as a result of a combination of structural and medical complications (Lazenby, 2008). The structural and anatomical features of Down syndrome which forms the basis of the orofacial, feeding and swallowing difficulties are:

Craniofacial features

- The principal skeletal craniofacial features include brachycephaly with a flattened occiput and decreased length and flattening of the cranial base as a result of vertical hypoplasia of the structures of the skull (Cohen & Winner, 1965; Gulliksen, 1973; Alonso, Naval, Hernandez, & Lucas, 1985; Farkas, Munro, & Kolar, 1985; Fischer-Brandies, 1988).
- The facial mid-third is underdeveloped; producing a hypoplastic maxilla with a high placed, short and narrow palate (Shapiro, Gorlin, Redman, & Bruh, 1967).
- Purdy, Deitz, and Harris (1987) stated that the frontal and paranasal sinuses are hypoplastic and Fischer-Brandies (1988) found the ethmoid bone to be retracted.

- Facial appearance is altered with short palpebral fissures, hypertelorism, low nasal bridge and a soft narrow nose, a high upper lip, and wide short low set ears (Moraes, Moraes, Dotto, Dotto, & Santos, 2007; Seabra, Macho, Pinto, Soares, & Andrade, 2008).
- Mandibular prognathism was found to be mild or marked relative to the maxilla (Farkas, Munro, & Kolar, 1985).
- Cleft lip, incomplete lip closure and hypotonic lip are present (Moraes et al., 2007; Oliveira, Paiva, Campos, & Czeresnia, 2008; Seabra et al., 2008).
- Soft-tissue features include a fissured and protruded tongue which gives the impression of being abnormally large on account of muscle weakness that often rests between the dental arches and an anterior and low position in the mouth (Oliveira et al., 2008; Suri, Tompson, & Atenafu, 2011). The tongue shows inaccurate and slow tongue movement (Moraes et al., 2007; Oliveira et al., 2008; Seabra et al., 2008).
- The tonsils and adenoids are enlarged and at times bifid uvula is also seen (Kavanagh, Kahane, & Kordane, 1986)
- Individuals with Down syndrome are mouth breathers, exhibit open bite and their orofacial muscles are hypotonic, there is an incomplete closure of the lips (Weijerman, Van Furth, Vonk Noordegraaf, Van Wouwe, Broers, & Gemke, 2008; Alió, Lorenzo, Iglesias, Manso, & Ramirez, 2011). It causes an imbalance in orofacial development which leads to malocclusion (Weijerman et al., 2008; Oliveira et al., 2010) and craniofacial malformations such as the hypoplasia of the midface (Moraes et al., 2004; Oliveira et al., 2010).

- Hypotonicity is associated with ligament laxity, which is easily visible throughout the body (Weijerman & Winter, 2010). It induces hyper flexible joints, which can compromise the periodontal ligaments.
- Excess of saliva on the labial commissure is also related to the muscle hypotonicity and can lead to irritation, cracking (angular cheilitis), aphthous ulcers and infectious conditions like candidiasis (Shore, Lightfoot,& Ansell, 2010; Areias, Sampaio-Maia, Guimaraes, Melo,& Andrade, 2011).

Occlusal anomalies

- A shortened palate in the anteroposterior dimension is seen as a result of underdevelopment of the midface bones (Shore et al., 2010).
- Anterior openbite, posterior crossbite and reductions in the maxillary arch are seen as a consequence of the variations in vertical and transversal dimensions. Thrusting of the tongue may also hamper eruption causing anterior open bite and thereby influencing the shape of the dental arch and the positioning of the teeth (Moraes et al., 2007; Oliveira et al., 2008).
- Malocclusion in Down Syndrome individuals increases with age and it happens due to craniofacial growth restriction, oral-motor dysfunction and generalized orofacial hypotonicity (Macho et al., 2014)
- Insufficient bone development associated with impaired facial muscles can lead to a weak lip closure strength, drooling and occlusal abnormalities (Oliveira et al., 2008, 2010)

Dental anomalies

- Abnormalities in the number (fewer), size (smaller) and morphology and the timing of their development (late dentition) are constant features of this syndrome. In the primary dentition, the most commonly absent teeth are lateral incisors, while in the permanent dentition, third molars, second premolars and lateral incisors, in this sequence, are the most frequently missing teeth (Moraes et al., 2007)
- Individuals with Down syndrome have complete tooth mineralization, delayed or abnormal sequence of tooth eruption (Fischer-Brandies, 1989; Oliveira et al., 2008) changes in the sequence of eruption (mainly of the temporary teeth), high incidence of impacted teeth (incisors and canines) and teeth agenesis (Moraes et al., 2007; Macho et al., 2014)
- Dental anomalies pertaining to number include hypodontia or partial anodontia, microdontia, enamel hypoplasia of deciduous teeth and oligodontia (Macho et al., 2014).
- Structural abnormalities include taurodontia, peg-shaped teeth, decreased root to crown ratio, decreased tooth size, altered crown shape (Bell, Civil, Townsend, & Brown, 1989) fusion and gemination. Canines are the most affected (Moraes et al., 2007; Macho et al., 2014).

Oral diseases

- There is also a high incidence of aphthous ulcers, oral candidiasis and acute ulcerative gingivitis (Amaral, Oliveira, & Eustaquio, 2007; Oliveira et al., 2007).
- The increased incidence of periodontal disease can be explained by the muscular hypotonicity and its consequences, dento-alveolar joint laxity, lack of understanding

of the needs of oral hygiene, impaired dexterity, compromised immune system, low T cells count and leukocyte dysfunction (Amaral et al., 2007; Oliveira et al., 2010; Khocht, Heaney, Janal, & Turner, 2011).

- Difficulty in gargling and swallowing, associated with a poor chewing ability reduces the natural teeth cleaning. Consequently, patients with this syndrome have halitosis, discomfort during chewing and early loss of permanent teeth (Macho et al., 2014).
- Literature reports a high incidence of esophageal atresia, and tracheoesophageal fistula in children with Down syndrome. In 2002, Bianca, Bianca, & Ettore confirmed 90 cases of esophageal atresia (0.03%) from 1991 to 1998 in 243,916 live births.
- Wallace in 2007 found among 57 adult patients with Down syndrome gastrointestinal issue such as celiac diseases, gastroesophageal reflux, achalasia, over nutrition, inflammatory bowel disease, cholelithiasis and unexplained constipation and diarrhea, where majority of malformations was likely to be present early in life.

Other diseases/defects

- Gastrointestinal diseases and constipation are present due to the low muscle tone and these difficulties in digestion could be aggravated by lack of chewing (Mercier & Poitras 1992). It is well documented in the literature that if left untreated, gastrointestinal issues can significantly complicate feeding.
- The presence of heart defects in about 40% of infants with Down syndrome, is another important risk factor which impacts an infant or child's ability to finish feeding and take enough volume to support health, growth and development. Depending on the type and severity of the heart defect, excessive perspiration, fast

and difficult breathing, vomiting, fatigue during feeding, inadequate food intake and limited weight gain can occur (Aumonier & Cunningham, 1983).

- Lastly, the risk for intrauterine growth retardation resulting in low birth weight in infants with Down syndrome and the combination of low birth weight and its sequelae, as well as low muscle tone, can result in early feeding problems in infants with Down syndrome (Venter, Christianson, Hutamo, Makhura, & Gericke, 1995).

Functional Consequences of Structural Anomalies

The functional manifestations of these structural abnormalities are indirectly related to the underlying pathology. The normal development of oral structure and its function is altered in infants and children with Down syndrome which in turn leads to the development of a compromised suckling, swallowing, and mastication (Fischer-Brandies, 1989; Stevenson & Allaire, 1991). The characteristic orofacial dysmorphology of Down syndrome represents an obstacle to the development of oral-motor coordination. Breastfeeding was found to be difficult in infants with Down syndrome due to hypotonia, a small mouth size, a less demanding personality, cardiac defects, and intestinal problems (Newman, 2006; Thomas, Marinelli, & Hennessy, 2007).

The development of a mature rotary chewing pattern is dependent upon sensorial stimuli, mainly from the periodontal dental receptors (Woda & Fontenelle, 1993). For children with Down syndrome the small size of the maxilla relative to the mandible makes it impossible for the teeth to interlock and the periodontal receptors are insufficiently stimulated to initiate the rotary lingual pattern of mastication. Orofacial dysmorphology also affects the stability of the mandible, which is the first condition for swallowing and biting food. The laxity of the temporomandibular joint ligaments also

facilitates the mandibular protrusion. This leads to one or more new resting positions to become established, which effectively traps the maxilla behind the mandible. Hence the child may develop a clenching or grinding habit in an attempt to find a position of comfort and subconsciously tries to eliminate occlusal interferences.

Mastication is impaired because of poor oral exploration and an inability to initiate, grade, or sustain oral patterns. Movement is arrhythmic with incoordination of the lips and mouth and excessive vertical opening (Light, 1995). This may lead to drooling and potential swallowing incoordination. In addition, the presence of an endognathic maxilla, an anterior open bite, and a reduced number of teeth may further decrease masticatory capacity. Muscular hypotonicity in the cervical region adds to the problem of swallowing as the neck is extended at rest (Woda & Fontenelle, 1993). The degree of difficulty experienced by each individual with Down syndrome is variable but is primarily due to hypotonicity of the muscles of mastication and facial expression, particularly of the tongue and lips. Children with Down syndrome are therefore obliged to displace actively the bolus backward by a primitive forward–backward movement of the tongue against the palate, and tongue thrusting is encouraged. The consequences of this abnormal function may be the development of a high narrow palate and an elongated tongue due to the preferential development of the longitudinal muscle fibers. The tongue is inefficient in lateralizing food for chewing particularly in the molar region (Gisel, Lange, & Niman, 1984).

Castillo-Morales, Crotti, Avalle, and Limbrock in 1982 summarized the consequences of orofacial pathology on oromotor and feeding aspects which has been depicted in table 2.4

Table 2.4

Development of orofacial pathology in infants with Down Syndrome (Source: Castillo-Morales et al., 1982)

Primary pathology	Secondary pathology
<p>Muscle hypotonia of orbicularis, zygomatic, masseter, temporalis and muscles of facial expression, reduced tonus of ligamentary apparatus of mandibular joint, disorders of immune system.</p>	<p>Lower lip everted, angle of mouth pulled down, upper lip inactive and pulled up, with hypoplastic lateral aspects and short frenulum, open-mouth breathing, chronic periodontitis, drooling, chapped lower lip and respiratory infections.</p>
<p>Hypotonic tongue protrusion and later active tongue protrusion, midline junction of tongue weak and protruding (lingual diastasis), excessive concavity of frontal two-thirds of tongue and weak frenulum.</p>	<p>Tongue protrusion or thrust during drinking, sucking pacifier, eating and speaking, relative macroglossia (rarely true macroglossia), dry, chapped tongue surface, upper and lower front teeth protrude, indistinct pronunciation/articulation.</p>
<p>Hypoplastic middle face, hypoplasia of maxilla in sagittal and transverse directions, reduced palatal height, hypotonic velum, sometimes submucous</p>	<p>Tongue protrusion also because of small oral cavity, maxillary sagittal growth and palatal height remain reduced, maxillary transverse growth progressively reduced, sometimes V-</p>

cleft palate and/or velum.	shaped palate, which seems to be high, velar insufficiency, sometimes contraction into bowl shape.
Reduced jaw angle and reduced total length of mandible, retarded dentition, microdontism, anodontism, hypodontism: aberrant teeth.	Progressive reduction in jaw angle, but less reduction in length of mandible, openbite with dento-alveolar components, protruding front teeth, pseudoprognathism, subluxation of jaw, retarded bite function, oral stereotypies.

Feeding Problems in Children with Down Syndrome

The infants with Down syndrome may experience problems with breast feeding, transition from breast to bottle feeding and to cup feeding, and from liquids to solids which can result in inadequate lip closure, poor chewing ability and choking (Kumin, 1994; Pipes, 1995; Cooper-Brown et al., 2008). Infants and children with Down syndrome face variety of problems such as 10-35% delay in acquisition of oral-motor coordination necessary for normal feeding milestones (Pipes, 1995); slow development of the ability to manipulate food in the fingers as well as in the use of feeding utensils; the failure to progress through a normal sequence of food textures and the refusal of certain food textures, particularly those of a hard texture; behavioral problems such as the refusal to swallow, the spitting out of food, or the retention of food or utensils in the mouth.

Literature even reports of silent aspiration as a problem among this population with liquid and semiliquid food reaching the bronchi. On a daily basis any aspiration can contribute to the incidence of pulmonary infection. Lack of coordination between

breathing and swallowing may induce either the aspiration of liquids or swallowing of air, followed by coughing or belching. Problems with swallowing have been described as being due to slow development of motor skills in the child (Cullen et al., 1981). However, such difficulties can often persist into adulthood and may have grave consequences for general health. In addition as a result of the early feeding problems in infants with Down syndrome and the emotional responses of parents to the often unexpected diagnosis of Down syndrome (Cunningham, 1996), early mother-infant attachment may be disrupted and breast feeding is often not possible (Van Dyke, Mattheis, Eberly, & Williams, 1995; Rynders & Horrobin, 1996; Gigacz, 2001).

Tests to Assess Feeding Problems

Feeding assessment is very important as it is a multifaceted heterogeneous disorder where every individual will have different difficulties which vary with severity. There are different feeding assessment scales that have been developed which tries to describe the child's feeding issue. Developmental pre-feeding checklist (Morris & Klein, 1987) is a tool that can be used for children of any age group but it mainly assesses the developmental skills emerging between birth to 24 months of age. It provides qualitative and quantitative description of feeding performance, brief history of the feeding problem and detects any abnormal oral pattern if present. It assesses feeding through bottle or breast, liquid by cup, semi-solid and solids.

Multidisciplinary Feeding Profile developed by Kenney, Casas, and McPherson in 1989 is meant for the patients who are dependent for feeding. It is a numerical rating scale developed to assess various components which includes physical/neurologic factors (posture, tone, reflexes, and motor control), oral-facial structure, oral-facial sensory

inputs, oral-facial motor function, ventilation/phonation and a functional feeding assessment. It assesses based on different kinds of feeding, i.e. spoon feeding, biting, chewing, cup drinking and straw drinking. This tool was found to be reliable based on the studies conducted by Judd, Kenny, Koheil, Miller and Moran (1989).

Oral-motor Feeding Rating Scale (Jelm, 1990) is an assessment tool which provides a concise picture of feeding problem in children and adults (1 year to adulthood). It also provides guidelines regarding the management of it. It has six point rating scale from normal to abnormal which assesses feeding by breast, bottle, spoon, cup, biting, chewing and straw drinking.

The Child Eating Behavior Inventory (CEBI) developed by Archer, Rosenbaum, and Streiner (1991) is to assess feeding problems and how the parent-child relationship is influenced. It is developed for children in the 2-12 years of age range and it is administered on the parents. It consists of 40 items and is rated on point rating scale with responses: never, seldom, sometimes, often and always. A cutoff score of 16% is indicative of having an eating problem.

Exeter Dysphagia Assessment Technique (EDAT) proposed by Selly, Flack, Ellis and Brooks (1989) and revised by Selly et al., (2001) is a non-invasive way to measure swallowing function to distinguish dysphagia because of sensory nerve, motor nerve or functional involvement. It uses diverse specialized equipments to document oral, respiratory and swallowing features.

Clinical feeding evaluation of infants (Wolf & Glass, 1992) assesses feeding difficulties among infants fed by bottle or breast, also assesses difficulty which may be present while spoon feeding and cup drinking.

The Neonatal Oral Motor Assessment Scale (NOMAS) given by Palmer, Crawley and Balnco (1993) is a checklist to evaluate the behaviors in categories of normal, disorganized and dysfunction of tongue and jaw movement in the age range of birth to 8 weeks. It was originally given by Braun and Palmer (1986) and revised by Case-Smith, Cooper, and Scala in 1989.

Behavioral pediatric feeding assessment scale developed by Crist, McDonnell, Beck, Gillespie, Barrett and Matthews (1994) contains 25 items which assesses child's feeding behavior and 10 more items with which parent's feeling about dealing with feeding problems of the child can be assessed. Also, there is a 5 point rating scale where parents have to tell how much they agree or disagree with the statements provided. Therefore, higher the score more problematic feeding behavior is present. This tool is found to be valid and reliable for representing child's and parents mealtime behavior.

Schedule for Oral Motor Assessment (SOMA) developed by Reilly, Skuse, Stevenson, and Mathisen (1995) is used to rate objectively oral-motor skills and recognize the areas of the dysfunction which might lead to feeding difficulties. It can be administered on children in the age range of 8-24 months. It covers 75-90 items which are scored pass or fail for each of the 6 food types administered. The items are divided into categories of jaw, lip and tongue movements and reactivity, acceptance, initiation, food loss/ drooling, sequencing and swallowing. They assess based on the different kinds

of food i.e., liquid (by breast/ bottle and/ or spouted cup, regular cup and straw), spoon feeding of puree, semi-solid and solid, and finger feeding of biscuit and dried fruits.

Children's eating behavior questionnaire developed by Wardle, Guthrie, Sanderson, and Rapoport (2001) is to assess a variety of aspect of the eating style based on eight dimensions i.e. responsiveness to food, enjoyment to food, satiety responsiveness, slowness in eating, fussiness, emotional over eating, emotional under eating, and desire for drinks.

Dysphagia Disorder Survey (DDS) - Pediatric developed by Sheppard (2002) is a screening instrument to assess feeding and swallowing function in children and adults with developmental disabilities (3-78 years).

Early feeding Skills (EFS) given by Thoyre, Shaker, and Pridham (2004) is a checklist for assessing infant readiness for tolerance of feeding and to profile the infants developmental stage according to feeding skills i.e. abilities to remain engaged in feeding, organized oral motor functioning, coordinate swallowing with breathing and maintain physiologic stability. There are many more feeding assessment tools which helps in assessing feeding difficulties in infants e.g. Infant Breastfeeding Assessment Tool (IBFAT), the Mother Baby Assessment Tool (MBA), the LATCH assessment tool, Preterm Infant Breastfeeding Behavior Scale (PIBBS), Systematic Assessment of the Infant at Breast (SAIB) and many more.

Intervention

The management of swallowing and feeding problems in infants and children with Down syndrome requires individualized treatment plans, which must be developed

with a clear understanding of normal oral-motor development and how the deficits observed in each child differ from normal development. Management plans take into account multiple interactive factors such as disease state, chronological and mental age of the child, physiological status, and psychological/behavioral factors. When working with infants and older children with dysphagia, a number of different professionals collaborate to provide an individualized treatment plan. The Speech-language pathologist could work with family members, neonatologist, pediatrician, gastroenterologist, occupational and physical therapists, nutritionist, nurse, radiologist, social worker, otolaryngologist, psychologist, surgeon, pulmonologist, dentist, respiratory therapist, lactation consultant. The team members will change, depending on the setting.

Studies Investigating Feeding Patterns in Children with Down Syndrome

Relatively few studies have exclusively examined the feeding difficulties faced by children with Down syndrome. Craig, Peter and Joyce in 1982 conducted case studies on seven children with Down syndrome with abnormalities in esophageal function. Three of these children had recurrent episodes of pneumonia from gastroesophageal reflux, and two of them had esophageal strictures. Though the other two cases revealed no evidence of gastroesophageal reflux they did show significant abnormalities in esophageal peristalsis. Case one of their study developed dysphagia more marked with solids than liquids. On later evaluation it was revealed that the case had hiatal hernia.

Infants with Down syndrome have physical features and medical issues that may complicate breast feeding. Torfs and Christianson in 1998 compared anomalies at birth in 2.5 million control infants to 2,894 infants with Down syndrome and found a higher incidence of hypotonia, cardiac defects, digestive system problems, eye cataract,

respiratory system defects, genital differences, anomalies of the extremities, urinary system problems, hydrocephalus, hernia, and cleft palate. In 2003, Pisacane, Toscano, Pirri, Continisio, Andria, and Zoli studied the prevalence and duration of breastfeeding among infants with Down syndrome and found breast feeding to be significantly lower with a mean duration of breast feeding days being 54 days in infants with Down syndrome as compared to 164 days in infants without Down syndrome.

In order to promote closer collaboration with families and speech-language therapists in early communication intervention Lewis and Kritzinger in 2004 investigated some experiences of a group of parents concerning the feeding problems of their infants with Down syndrome. A descriptive survey approach was implemented to collect quantitative data from 0-3 month's old infants with Down syndrome. The results revealed the types of feeding problems and associated conditions occurring in the infants with Down syndrome; the type of feeding methods used; the assistance received during the feeding problems; the emotions experienced in relation to the feeding problems in their infants as well as suggestions made by the participants regarding the management of early feeding problems. Twenty infants with Down syndrome were assessed via parental questionnaire. Numerous feeding problems occurred in these infants and the variety of emotions experienced by the participants indicated that feeding therapy and counseling were important. The feeding problems observed in these infants were weak lip seal, an uncoordinated suck swallow-breathe pattern, problems with positioning for feeding and severe fatigue all of which were mainly associated with reduced muscle tone. The potential consequences of these difficulties lead to problems such as coughing, choking, aspiration, vomiting and weight loss. As a result of the different conditions underlying

the feeding problems, a comprehensive approach, which included therapeutic and medical intervention, was necessary. In order to provide effective early communication intervention services from birth, the results indicated that the knowledge, skills and sensitivity of speech language therapists to identify caregiver needs, to provide appropriate family-focused intervention and to make recommendations regarding the management of feeding problems in infants with Down syndrome is of great importance.

Gisel, Lange, and Niman (1984) conducted video recordings of 4- to 5-year-old children with Down syndrome eating a standardized meal. The study included twenty-six children with Down syndrome, where 14 of them was aged 4 years \pm 2 months (8 males, 6 females) and 12 of them was aged 5 years \pm 2 months (6 males, 6 females). This age group was compared with those children of a study with normal 4-and 5-year-old children by Schwartz, Niman, and Gisel, (1984). Each child who was sitting on a chair was given twelve raisins, six bites of graham cracker, and six small spoonful of unsweetened applesauce. The investigator sat on the floor in front of the child to see into the child's mouth and the video camera was placed to the left of the investigator. The chewing cycles were videotaped. Three sets of data were obtained from the videotapes, 1. the number of chewing cycles needed to swallow each mouthful of food, 2. the time (in seconds) from the moment food was placed in the mouth until the last swallow, 3. the average cycle length for each mouthful of food (time/ cycle ratio). Their study revealed that the time/ cycle ratio was different for all foods tested, except for applesauce. Tongue protrusion is marked in children with Down syndrome and food is held longer in the mouth when compared to control subjects, whether solid, semisolid, or purée. This was attributed to the delay in the initiation of oral movement in children with Down

Syndrome and partly due to pauses within the masticatory cycle. They also observed that children with Down syndrome exhibited a reluctance to chew and preferred sucking of food. Abnormal chewing cycles was observed as a consequence of approximating a dental contact despite a marked malocclusion. This involves a degree of mandibular protrusion which, along with hyper laxity of the temporomandibular joint ligaments, results in chronic luxation of the joint. The study also revealed that the decreased masticatory efficiency could be due to a loss of sensory input and may decrease the pleasurable aspects of eating. Hence children with Down syndrome who swallow their food whole may tend to overeat in order to compensate for the loss of gustatory input, thereby leading to the development of bulimia and obesity in children with Down syndrome.

Frazier and Friedman in 1996 conducted a retrospective study in order to describe the swallowing function in children with Down syndrome who were referred to the Swallowing Disorders Clinic of The Children's Hospital (TCH) between 1989 and 1994, Denver as well as to identify the possible factors that may influence respiratory health in this population. The data was extracted from a standardized database, where 19 children in the age range of 3 months to 11 years were selected. The subjects were predominantly male, that is sixteen males and three females. In order to obtain details on respiratory, cardiac and gastrointestinal history and details of eating method, a retrospective chart review was completed on all 19 patients. Videofluoroscopic modified barium swallow (VMBS) was used to understand swallowing function and dysfunction in children with Down syndrome. The speech-language pathologist, the occupational therapist and the dietitian conducted a pre-video fluoroscopic assessment before VMBS was administered

on each child. This assessment compiled details of a parent/ caregiver interview, an oral motor, oral sensory and feeding assessment, analysis of the child's postural control and a nutritional evaluation, following which VMBS was administered. The food textures presented and the method of administration were selected to match as closely as possible to those being consumed by the child on a daily basis and it varied depending on the child's developmental level and their eating and swallowing ability. Five aspects of swallow function were evaluated during the VMBS: (1) oral preparation, (2) reflex initiation, (3) pharyngeal clearing, (4) aspiration and (5) cricopharyngeal screening. Their findings suggested that their oral phase may be affected by oral hypersensitivity which can interfere with their acceptance of textured foods. Cluster of behaviors associated with oral sensory dysfunction was observed. Rejection of age appropriate food textures, reduced acceptance of food tastes, temperatures, or smells, picky eaters, aversive or exaggerated response to touch in or around the mouth, hyperactive gag response, aversion to brushing teeth, lack of age-appropriate mouthing of toys/hands was the cluster of behaviors observed for oral hypersensitivity. Poor or no awareness of food on lips, slow registration of food in mouth, pocketing of food, stuffing of mouth indicated the cluster of behaviors associated with oral hyposensitivity. In 16 children evaluated a disordered pharyngeal phase, with aspiration in 10 of the 19 children studied was identified. Aspiration observed were silent for eight of these 10 children which did not correlate with the severity of impairment that is present in the oral phase. Besides considering other factors such as sore throats, bronchitis, pneumonia, upper airway obstruction, immunodeficiency, gastrointestinal reflux, congenital heart disease, hypoplastic lungs, this study also suggested aspiration as an additional factor that can predispose and

increase the risk of developing respiratory illness. The marked hypotonicity seen in children with Down syndrome was postulated to be a possible cause for the pharyngeal incoordination and subsequent aspiration. The study also revealed the difficulties faced by these children with advancing textures, reduced acceptance of food tastes, temperatures and smells which could be attributed to poor control of tongue movement which resulted in gagging and rejection of age appropriate textured food. Besides presence of decreased jaw strength, low muscle tone, and delays in acquisition of tongue movement affected efficient chewing and swallowing. The small oral cavity, laxity of the supportive musculature and enlarged tonsils also added on to the difficulties in swallowing solids.

Spender, Stein, Dennis, Reilly, Percy, and Cave (1996) explored in detail the nature of the feeding patterns in a sample of young children with Down syndrome who exhibited differences in oral-motor function from typically developing children. The multimethod approach they implemented included the assessment of oral-motor function which was done using Schedule for Oral Motor Assessment (SOMA, Skuse, Stevenson, Reilly, & Mathisen, 1995). For mother-child interaction during feeding, Feeding Interactional Schedule (FIS, Reilly, Skuse, Mathisen, & Wolke, 1995) was used; in addition anthropometric and developmental assessment to assess child's growth and general development, a history of the child's feeding and growth and assessment of psychological factors for assessing parent's own mental health and marital relationship were carried out. The investigators examined oral-motor function and feeding behaviors in 14 children with Down syndrome ranging in age from 11 months to 3 years. Performance was compared to 58 children matched according to mental age. The study

showed that young children with Down syndrome had significant impairments in oral-motor function. These impairments being just not a consequence of developmental delay but represent an aberrant developmental path, which appears to be specific to the syndrome. Oral-motor problems in the Down Syndrome subjects included delayed initiation and poor coordination and sequencing of oral motor movements, difficulty grading jaw movements for chewing, weak lip closure, and weak and reduced tongue movements.

Kumin and Bahr (1999) studied the patterns of feeding, eating and drinking in young children with Down syndrome with oral motor concerns. Thirty children with Down syndrome from 8 months to 4 years 11 months were taken for evaluation of oral motor skills related to feeding, eating and drinking. Parents were made to complete an oral motor questionnaire and an interview based on the questionnaire was conducted. All parents were made to observe, participate and provide feedback in the evaluation process. A battery of oral- motor behavior observations was developed, which was based on assessments by Mackie (1996) and Jelm (1995). Each evaluation included detailed observation of postural stability, position of the oral structures at rest, spoon feeding, chewing, bolus formation and drinking. These children were given age appropriate foods and liquids. They were also given a wide variety of food textures. Each evaluation lasted between 2 to 3 hours and included observations of the child, observations of the parent feeding the child, as well as parent-professional consultation and training. Postural concerns were demonstrated by approximately half of the children. The children with Down syndrome in the study were generally found to have symmetrical patterns in the areas of posture and jaw movements. The study revealed that different degrees of

hypotonia were evidenced in different oral structures. Low muscle tone in the lips was demonstrated in 44% of the children, while low muscle tone in the tongue was exhibited in 80% of the children. 71% of the children investigated were found to have maintained open mouth posture during rest, which could be related to the low muscle tone in the lips, jaw instability, loose ligaments in the temporomandibular joint or a mouth breathing pattern. The tongue was protruded during spoon feeding in 86% of the children. On foods that required chewing nearly 93% of the children protruded the tongue during swallowing, while 86% of the children demonstrated tongue protrusion during drinking. These children also demonstrated limited tongue retraction during swallowing. Nearly 60% of the children formed an adequate bolus while eating soft foods. The authors also found that young children with Down syndrome faced problems in sensory awareness and feedback which impacted feeding, eating, drinking and speech production. The result supported many of the anatomical and physiological findings that have been reported in literature regarding the associated characteristics of children with Down syndrome.

Hennequin, Allison, and Veyrune in 2000 conducted a cross-sectional study to describe the oral health problems in a sample of children with Down syndrome compared to their siblings. Data was collected using the Oral Assessment – Down Syndrome (OADS, Allison & Hennequin, 2000) questionnaire, which is a validated French language evaluation tool to be completed by the child's caregiver. The questionnaire was anonymously answered by parents who were attending a national meeting. They returned the replies of the questionnaire for their child with Down syndrome and for the sibling closest in age by post. Data concerning 204 individuals with Down syndrome (103 males, 101 females; mean age 9.6 years) and 161 of their siblings (80 males, 81 females; mean

age 11.8 years) were returned. The association between Down syndrome status, age, and oral health was analyzed in the study. There was a higher frequency of oral health related problems in those children with Down syndrome with respect to clinical signs and symptoms, function, disability, and development. The functional impairment experienced by children with Down syndrome impacted all of the oral functions during maturation. Suckling, swallowing, and chewing were delayed and breathing coordination remain impaired. The OADS was able to reflect such problems clearly. The prevalence of teeth grinding and tongue thrusting was found to be high for children with Down syndrome. The feeding pattern observed in children with Down syndrome was found to be an intermediate condition between a primary suckle–swallow pattern and full rotary chewing. Ineffective and depressed control of food between the jaws, lips, and tongue was observed. For the same reasons, children with Down syndrome were shown to spill their food during meals. Children with Down syndrome chewed food for short periods due to lack of a mature masticatory pattern. This high percentage can be linked to another observation found in the study that 53% of young children with Down syndrome and 40% of older children with Down syndrome were constipated. Gastrointestinal diseases could also be aggravated by lack of chewing (Mercier & Poitras, 1992). As the child grew older the prevalence of chewing problems did not improve, there was an increase in the prevalence of bleeding gums and mouth breathing with age, and tongue protrusion subsided with age. Most parents rated the general and overall oral health of their children with Down syndrome high, worse than for their siblings. They concluded that this group had a high level of functional and dysfunctional oral health problems some of which persisted into adulthood.

Field, Garland, and Williams (2003) conducted a study to examine the correlation of specific childhood feeding problems and possible predisposing factors for feeding problems. Through an interdisciplinary team they examined the patient notes of 349 children with and without intellectual disability. The review of records conducted revealed that each participant of the study revealed to have at least one or more of the five functionally defined feeding problems: food refusal, food selectivity by type, food selectivity by texture, oral motor delays, or dysphagia. The prevalence of predisposing factors for these feeding problems on examination revealed that the predisposing factors included developmental disabilities, gastrointestinal problems, cardiopulmonary problems, neurological problems, renal disease and anatomical anomalies. The frequencies of predisposing factors varied based on the feeding problem. The study revealed differences in the prevalence of the five feeding difficulties among the children with three different developmental disabilities: autism, Down syndrome and cerebral palsy. The authors discovered a significantly higher prevalence of oral-motor problems, swallowing difficulties and texture selectivity in the 26 children with Down syndrome than in the other children without Down syndrome. They suggested that children with Down syndrome had the ability to chew but refused to do so as a result of learned aversions to specific textures that prompted unpleasant experiences such as gagging and vomiting.

Sooben in 2012 reviewed seven studies from seven different countries spanning over 30 years in order to examine the factors that influenced breastfeeding among mothers of infants with Down syndrome. The review indicated that these children with Down syndrome presented with a range of difficulties in breast feeding during the first

few days of life, as a consequence of anatomical, facial, and structural abnormalities associated with Down syndrome. A high rate of breastfeeding initiation was associated with the involvement of professional breastfeeding support. The barriers to breastfeeding initiation were linked to the infants' craniofacial features and medical issues leading to an inability to feed. The medical condition took a toll over breastfeeding needs and the maternal infant feeding decision. The studies also revealed delay in introducing solid food compared to children without Down syndrome. Besides those infants who were breast fed for 6 months, presented with a reduced susceptibility to acute leukemia.

Mohamed, Alhamdan, and Samarkandy (2013) directed their study in order to understand the dietary practices and physical activity in children with Down syndrome and their siblings in Saudi Arabia. The study group consisted of Down syndrome boys and girls (n=108) clinically and/or cytogenetically proven to be ailing with Down syndrome aged 5-12 years. In order to ensure quite similar environmental backgrounds, they included their healthy siblings, closest in age to the children with Down syndrome as a control group (n =113). Some of the siblings included in the study were twins. All the children with Down syndrome included in their samples were living with their parents and had at least one sibling; where all siblings of Down syndrome children were living in the same house. The observer personally interviewed 13 illiterate mothers and 10 mothers with elementary education who were not able to fill the questionnaire. The history of infant feeding among children with Down syndrome and their siblings was obtained including type of feeding and duration of breast feeding. The observer recorded the nutritional feeding problems of children with Down syndrome and their siblings, which included chewing and swallowing difficulties, difficulties in using utensils, food rejection

and refusal. Information regarding physical activity status for the children with Down syndrome and matched siblings were collected from their parents. In order to evaluate dietary habits food frequency questionnaire was used. Parents were interviewed in order to obtain information regarding the daily details of food consumed by their children. The results of the study indicated that during infancy period, nearly half of the breast-fed children with Down syndrome were fed for a duration of less than 6 months besides 36.4% of the children with Down syndrome were bottle fed, compared to only 5.5% of the normal siblings. This indicated the difficulties the children with Down syndrome had in transition from breast and bottle feeding to cup feeding, and from liquids to solids. They found that the percentage of children with Down syndrome experiencing dietary difficulties was significantly higher when compared to their siblings. Concerning physical activity, 73.1% of Down syndrome children did not exercise as compared to 44.2% of the control siblings.

To sum, the review of the existing literature revealed that feeding which is a crucial process for the overall growth and development of the child, is affected in children with Down syndrome. These deficits could arise due to the structural abnormalities which affect the normal development of oral structures and its functions in infants and children with Down syndrome which in turn leads to the development of a compromised suckling, swallowing, biting and chewing. As a sequela of various anomalies like cardiac defects, eye cataract, hypotonia, respiratory system defects, anomalies of the extremities and other health related issues, studies have revealed a variety of problems faced by infants with Down syndrome that could interfere with breastfeeding and delay in the suckling ability. As the child grows older problems with

drooling, biting and mastication, difficulty in progressing at the normal sequence in manipulating food in fingers and in the use of feeding utensils; difficulty in transitioning to varied textured table food and the refusal of certain foods particularly those of a hard texture; behavioral problems such as the refusal to eat and swallow food, overeating food, eating a limited variety of food, spitting out of food, or the retention of food or utensils in mouth are seen. If these problems are left unattended, the feeding difficulties in children with Down syndrome may have significant long-term perennial health consequences, including growth deficits, insufficient nourishments and reduced performance on academic and cognitive tests. Further these could also have a negative impact on the quality of life of the child and on his/her parents/caregivers as well.

Although various studies have evaluated the nature and extent of feeding problems, most of them are retrospective evaluation of the cases, thereby hindering a direct observation, assessment and evaluation of children with Down syndrome. Further most of these studies have been on infants focusing on breastfeeding difficulties seen in them. It is not known whether children overcome these deficits as they grow older with intervention. Also studies exploring different aspects of feeding and the phases of swallowing are also limited. Thus a need was felt to assess the feeding patterns in children with Down syndrome in the age group of 2-7 years. Keeping this perspective in view, the present study was planned.

Chapter 3

Method

The present study aimed at investigating the feeding problems, if any, in children with Down syndrome in the age group of 2-7 years and to compare it with the chronologically matched typically developing children. The study also aimed at assessing the oromotor problems if any, developmental changes if any, effects of gender and the effects of intervention on feeding skills. The study was undertaken in the following phases:

Phase I: Construction of a questionnaire to assess feeding skill.

Phase II: Administration of the questionnaire on the participants.

Phase III: Assessment of test-retest reliability.

Phase I: Construction of questionnaire to assess feeding skills

As a part of construction, the following research steps were undertaken:

Step 1: Development of a questionnaire to assess feeding problems

This step involved the development of a questionnaire to assess feeding problems faced by children with Down syndrome. This was prepared by collating information from the literature and based on the complaints concerning feeding received from the clients registered in the Special clinic for motor speech disorders, Department of Clinical Services, All India Institute of Speech and Hearing, Mysuru. The questions focused on the physical problems faced by the children during feeding and swallowing. There were

questions which focused on orosensorimotor issues, general feeding issues, feeding history, modifications during feeding and the different phases of swallow. The questions were grouped under four sections: I. Demographic data and general history, II. Craniofacial and orosensory assessment, III. Feeding history and IV. Assessment of different phases of swallow. The details of each of the section are given below:

- I. Demographic data: This section included the personnel details of the child, medical history (prenatal, natal, post natal history), results of different evaluations (speech and language evaluation, physio/occupational therapist evaluation, psychology evaluation) and general history.
- II. Craniofacial and orosensory assessment: This section was divided into three subsections:
 - a) Craniofacial assessment: Questions such as ‘Is the jaw size abnormal? Is the tongue size abnormal?’ etc. were included.
 - b) Oral hygiene: Questions such as ‘Does the child have poor oral hygiene? Is there plaque and debris accumulation?’ etc. were included.
 - c) Orosensory assessment: Questions such as ‘Is it difficult for the child to perceive the sensation of light touch on lips? Is it difficult for the child to perceive the temperature variations on the tongue?’ etc. were included.
- III. Feeding history: This section was also divided into three subsections
 - a) General feeding issues: Questions such as ‘Do you need to force the child to eat? Does the child have any food allergy?’ etc. were included.

b) Feeding history: Questions such as ‘Did the child have difficulty with feeding from the breast? Did the child have difficulty in eating independently with spoon?’ etc. were included.

c) Modifications during feeding: Questions such as ‘Is there any modification made to the utensils used for feeding? Is any alteration made to temperature of the food item to suit child’s needs?’ etc. were included.

IV. Assessment of different phases of swallow: This section included questions to assess the different phases of swallow. It was divided into three sections: a) Oral preparatory phase, oral phase, b) Pharyngeal phase and c) Esophageal phase of swallow. Questions such as ‘Is it difficult for the child to form a bolus? Is aspiration /choking during liquid intake seen?’ etc. were included.

Each statement in the IInd, IIIrd and IVth section was accompanied with response choice of “no” (a score of zero) or “yes” (a score of 1). A 5 point rating scale was also prepared to assess the severity of the feeding problems in the IVth section of the questionnaire at the end of every phase of swallow. The severity rating varied from 0-4 where, ‘0’ represented no difficulty at all; ‘1’ represented mild feeding difficulty; ‘2’ represented moderate feeding difficulty; ‘3’ represented moderately severe feeding difficulty; and ‘4’ represented severe feeding difficulty. At the end of the IVth section a 5 point rating scale was inserted to assess independently the overall severity of feeding problems faced by the child by the main feeder and the investigator.

Step 2: Content validity check

The content validity of the questionnaire and the rating scale was assessed by obtaining the feedback from three experienced speech-language pathologists. They were asked to judge the appropriateness of the questions included and the rating scale used. The feedback was collected using a 3 point rating scale ranging from the contents are not very valid (score 0) to all the contents are valid (score 2).

As per the ratings obtained from the three experienced speech-language pathologists, the different sections of the questionnaire were given a score of 2 which indicated that the contents were valid. However few suggestions were provided by the speech-language pathologists to remove the oromotor assessment and replace with Com-DEALL, Checklist for Assessment of Oro-motor skills in Toddlers (Archana & Karanth, 2008) and to include questions on oral hygiene. After the content validation, it was found that there was a need to include and delete a few questions under the different section of the questionnaire for obtaining a better understanding of the nature of the problem. In the demographic and general history section, three questions were included. Fifteen questions on craniofacial abnormalities and three questions on oral hygiene under the craniofacial and orosensory assessment section of the questionnaire were added. In the section on feeding assessment two questions from general feeding issues were removed due to lack of clarity. Further in the section involving phases of swallow the oral preparatory and oral phase of swallowing was merged as a single subsection as clear cut discrimination between both was phases was not present, and most questions were overlapping.

Step 3: Pilot study

A pilot study was carried out on 10 children with Down syndrome and on 10 typically developing children. The questionnaire was administered on two parents/caregivers of children with and without Down syndrome in different age groups between the age range of 2-7 years. A pilot study was carried out in order:

- a. To familiarize with the administration procedure
- b. To check if any relevant questions were missed out
- c. To check whether the procedure that would be adopted during data collection was appropriate and
- d. To find the approximate time taken in collecting data from one child

A video recording of the child's feeding skill was carried out in order to facilitate a better view of the feeding problems presented by the children as well as for later analysis. In addition they were also questioned about any other additional difficulties they faced while feeding their children. The responses obtained were documented. At the completion of the section four of the questionnaire the parents/caregivers were asked to rate the overall severity of their child's feeding and swallowing for eating and drinking separately on the 5 point rating scale. After the pilot study no changes were incorporated in the questionnaire or the procedure of data collection.

Step 4: Finalization of the questionnaire

The final version of the questionnaire was prepared after incorporating the feedback obtained from the professionals during the content validation and the responses obtained during the pilot study. The final form of the questionnaire had four sections covering demographic data and general history, craniofacial and orosensory assessment,

feeding history, assessment of different phases of swallow. The final version of the questionnaire has been provided in the Appendix.

Phase II: Administration of the questionnaire on the participants

The final version of the questionnaire was administered on 17 children with Down syndrome and 47 typically developing children in the age range of 2-7years. The details of the participants have been provided below

Participants

Seventeen children with Down syndrome (8 females and 7 males) in the age range of 2-7years, who reported to the Department of Clinical Services, All India Institute of Speech and Hearing, Mysuru, participated in the study. They were diagnosed as 'Delayed Speech and language with Down Syndrome' by a qualified team of professionals including speech-language pathologist, pediatrician and a clinical psychologist. The degree of Intelligence quotient ranged from near normal intelligence to moderate retardation. These participants were further subdivided into two groups based on their age. There were 9 children in the higher age group (5-7 years) and 8 in the lower age group (2-5 years). They constituted the clinical group. All the children included in the study were enrolled in an intervention program such as speech-language therapy, physio/occupational therapy and preschool. Based on the duration of intervention the participants were sub grouped into those who had attended the intervention program for less than 2 years and those who had attended for more than 2 years. Children with a history of co-morbid problems such as autism, cerebral palsy and other developmental disabilities were excluded. Two children had hyperthyroidism, six had atrial septal defect,

two had chronic upper respiratory tract infections, six had respiratory issues such as asthma and mouth breathing, four children had behavior problems, three had myopia, one child had squint and one child had mild hearing loss in the left ear. All the participants belonged to the middle class socioeconomic status which was ascertained using the NIMH socioeconomic status scale developed by Venkatesan (2011). The scale has sections such as occupation and education of the parents, annual family income, property, and percapita income to assess the socioeconomic status of the participants. Their oromotor abilities were assessed using Com-DEALL Oro Motor Checklist (Archana & Karanth, 2008). Three domains i.e. Jaw movement, tongue movements and lip movement with a total of 24 questions were considered for evaluation. Responses were rated on a three point rating scale where '0' signified absent, '1' signified only spontaneously present and '2' signified consistently present. The demographic details of the clinical group have been provided in the table below:

Table 3.1

Demographic details of the clinical group.

SNo	Participant No.	Chronological Age	Gender	Degree of Retardation	Associated Problems
1	P1	2;10	Male	Below average intelligence	Hyperthyroidism, atrial septal defect.
2	P2	3;2	Female	Average Intelligence	Atrial septal defect.
3	P3	4;0	Female	Below average intelligence	None
4	P4	4;5	Male	Below average intelligence	Behavior problems
5	P5	4;8	Female	Mild	None
6	P6	5;0	Male	Mild	Seizures, history of Chronic Respiratory Upper Tract

					Infection, behavior problems
					hyperthyroidism
7	P7	5;0	Male	Below average intelligence	Atrial septal defect, Chronic Upper Respiratory Tract Infection
8	P8	5;0	Female	Below average intelligence	Respiratory issues such as asthma
9	P9	5;5	Male	Moderate	Behavioral problems, respiratory issues such as asthma and mouth breathing.
10	P10	5;5	Female	Mild	Myopia
11	P11	5;6	Male	Near Normal Intelligence	Respiratory issues such as asthma.
12	P12	5;11	Female	Mild	Atrial septal defect, respiratory issues such as asthma

13	P13	6;1	Female	Borderline Intelligence	Myopia
14	P14	6;4	Male	Mild	Behavioral problem
15	P15	6;9	Female	Moderate	Squint, Mild hearing loss in the left ear, history of asthma, atrial septal defect
16	P16	7;0	Male	Mild	Myopia, behavioral problem.
17	P17	7;1	Male	Mild	Atrial septal defect, respiratory issues such mouth breathing.

A group of forty seven typically developing children (20 females and 27 males) matched for age and socioeconomic status were selected and they comprised the control group. There were 17 children in higher age group (7 females and 10 males) and 30 in the lower age group (13 females and 17 males). Their oro-motor abilities were also assessed by administering the Com-DEALL, Checklist for Assessment of Oro-motor skills in Toddlers (Archana & Karanth, 2008). The following criteria were adhered to while selecting the participants of the control group:

1. Participants with no history of neurological, oromotor, communicative, cognitive, or sensorimotor, and academic impairment were included. This was ensured using the 'WHO Ten-question disability screening checklist' (Singhi, Kumar, Malhi, & Kumar, 2007).

2. Participants who had age adequate language abilities were selected which was ascertained using Assessment Checklist for speech-language domain (Swapna, Jayaram, Prema, & Geetha 2010).

All ethical standards were met for participant selection and their participation. Prior to testing, a written consent was obtained from the parents of the participants after explaining the purpose of the study. Participants belonging middle socio-economic statuses were included which was ascertained using the NIMH socioeconomic status scale developed by Venkatesan (2011).

Procedure

The testing was carried out in a relatively noise free environment with minimum distractions. Each child was tested individually. A rapport was established with the mother/caregiver. The purpose of the administration was explained. The demographic data was obtained initially. The WHO Ten-question disability screening checklist and the Assessment Checklist for speech-language domain were administered on the typically developing children. The oro-motor abilities will be assessed by administering the Com-DEALL, Checklist for Assessment of Oro-motor skills in Toddlers (Archana & Karanth, 2008) followed by the final version of the questionnaire developed was administered. A video recording of the child's feeding skill was carried out in order to facilitate a better

view of the feeding problems presented by the children as well as for later analysis. The child was given different items to eat and drink (e.g., Biscuit, banana, water etc.) to permit a first-hand observation of the feeding skills. The feeding problems faced by the child were noted. At the completion of the section four of the questionnaire the parents/caregivers were asked to rate the overall severity of their child's feeding and swallowing for eating and drinking separately on the 5 point rating scale. The time taken to administer the questionnaire was approximately 45 minutes. On the whole to administer all the necessary protocols, the time taken was approximately 60 minutes. Positive reinforcements like verbal and social reinforcements were provided to maintain the interest and motivation of the child throughout the test administration.

Phase III: Assessment of test-retest reliability

To assess the test-retest reliability the questionnaire was administered again on ten participants selected randomly from the clinical group and the control group after one week of their initial responses. 31.25% of the overall sample from both groups was considered for test-retest reliability.

Analysis

The scores obtained from each participant in the questionnaire in both the groups were totaled. A total score on the feeding problems for each phase of swallow were obtained.

Statistical analysis

The total scores obtained in section IV and the sub section scores from all the participants were fed to the computer for statistical analysis. SPSS version 20 software was used for the statistical analysis. Descriptive statistics was used to obtain mean, median and standard deviation of scores obtained on the questionnaire for both the groups. Shapiro–Wilk test was administered to check for normality. Since the data did not follow a normal distribution and due to high standard deviation, nonparametric tests were used for statistical analysis. Descriptive statistics to obtain mean, median and standard deviation for both the groups. Mann-Whitney U test was used to compare the clinical group and control group and to measure the influence of different independent variables. Friedman’s test and Wilcoxon Signed Rank Test was used to measure the influence of different independent variables in the clinical group. Cronbach’ alpha to determine the test-retest reliability. The results obtained from all the above statistical measures have been presented and discussed in the next chapter.

Chapter 4

Results and Discussion

The present study aimed at assessing the feeding problems, if any, in children with Down syndrome. The specific objectives of this study were to compare the feeding problems in children with Down syndrome with that of the typically developing children, to compare the feeding abilities between the lower vs. higher age group of children in both groups and to compare the feeding difficulties across gender (males vs. females) in both the groups. The study also investigated the influence of intervention on feeding problems in children with Down syndrome.

A questionnaire was developed which focused on the physical problems faced by the children during feeding and swallowing. The questions were expected to elicit problems related to oral, pharyngeal and esophageal phase of feeding and swallowing (section IV of the questionnaire). There were also questions which focused on craniofacial and orosensory issues, general feeding issues, feeding history and modifications during feeding (section I, II, III of the questionnaire).

The questionnaire was administered on a total of 64 participants. Two groups of participants were considered. The clinical group consisted of 17 children with Down syndrome and the control group consisted of 47 typically developing children in the age range of 2-7 years. The scores obtained from the section IV of the questionnaire were totaled, tabulated and later subjected to statistical analyses using SPSS software version 21. Shapiro–Wilk test was administered to check for normality. Since the data did not

follow a normal distribution and due to a high standard deviation, nonparametric tests were used for statistical analysis. The following statistical procedures were carried out:

- Descriptive statistics to obtain mean, median and standard deviation for both the groups.
- Mann-Whitney U test, Friedman's test and Wilcoxon Signed Rank Test to compare the influence of different independent variables.
- Cronbach' alpha to determine the test-retest reliability

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

- I. Test retest reliability
- II. Comparison of feeding skills between the clinical and the control group
- III. Comparison of feeding abilities between age groups of children in both the groups
- IV. Comparison of feeding abilities across gender of both the groups
- V. Influence of intervention on feeding abilities in the clinical group

I. Test-retest reliability

The test retest reliability was determined for 10 children in the clinical group and 10 children in the control group. The alpha values for the scores obtained on the different subsections of phases of swallow for the clinical group was found to be strong (Oral phase (Solid)=1.00, Oral phase (Liquid)=0.99, Pharyngeal phase (Solid)=0.99, Pharyngeal phase (Liquids)=0.98, Esophageal phase=1.00) which indicated significantly high test-retest reliability. The alpha values for the scores obtained on the phases of

swallow for the control group was also found to be strong (Oral phase (Solid)=0.97, Oral phase (Liquid)=1.00, Pharyngeal phase (Solid)=1.00, Pharyngeal phase (Liquids)=1.00, Esophageal phase=1.00) which indicated significantly high test-retest reliability.

II. Comparison of feeding skills between the clinical and the control group

The clinical group (children with Down syndrome) was compared with the control group (typically developing children) for the total scores obtained on the phases of swallow (section IV of the questionnaire). A higher score on this checklist indicated greater feeding difficulties. The mean and standard deviation obtained have been depicted in table 4.1. On comparison, it was seen that the total mean scores for all the three phases (oral, pharyngeal and esophageal phases) were higher for the clinical group than for the control group. On comparison of the mean scores for each phase (oral, pharyngeal and esophageal phases) it was seen that these were also higher for the clinical group than for the control group. The results of Mann-Whitney U test revealed that there was a statistically significant difference between the two groups in all the phases of swallow. The $|z|$ values have been depicted in table 4.1. The performance of both the groups across the phases of swallow has been depicted in figure 4.1.

Table 4.1

Mean±standard deviation (SD) and |z| value of both the groups on phases of swallow.

Phases of swallow	N	Clinical Group	N	Control Group	 z value
Oral phase (Solids)	17	6.76±4.75	47	0.34±0.75	5.62*
Oral phase (Liquids)	17	3.58±4.13	47	0.06±0.24	4.79*
Pharyngeal phase (Solids)	17	4.76±2.88	47	0.34±0.81	6.17*
Pharyngeal phase (Liquids)	17	1.17±1.70	47	0.04±0.20	4.59*
Esophageal phase	17	0.64±1.05	47	0.08±0.40	2.85**
Total score	17	18.17±13.55	47	0.89±1.50	6.18*

Values are given as Mean±SD, * p < 0.001, **p<0.01

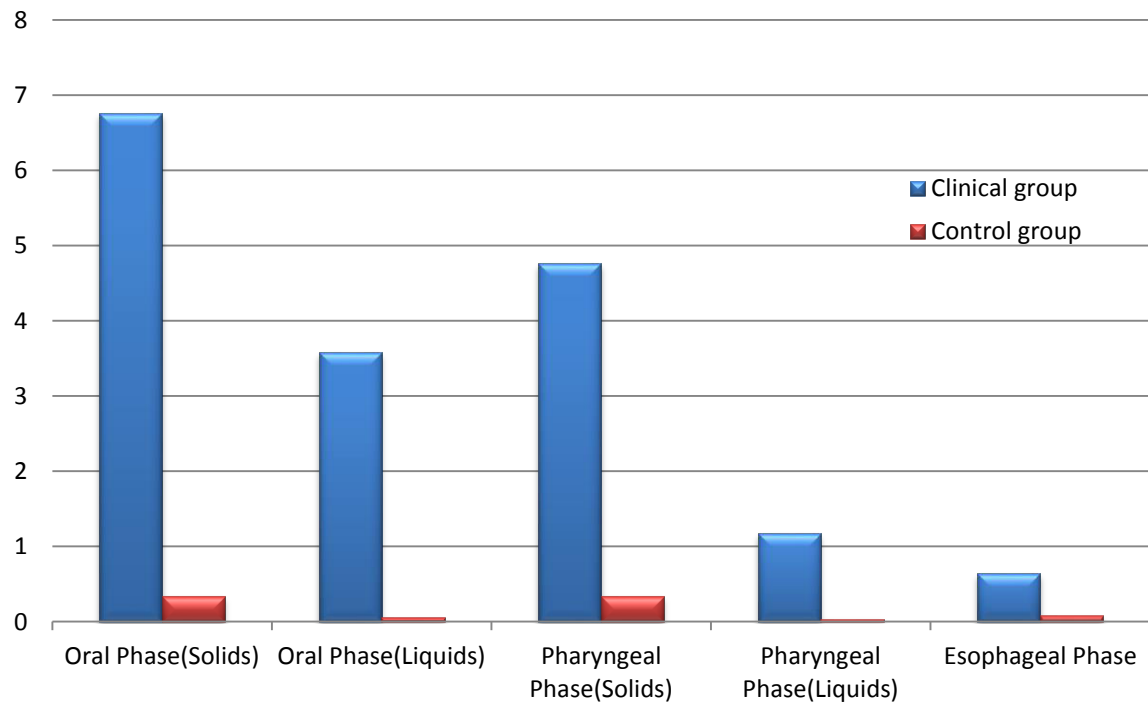


Figure 4.1. Mean scores on the phases of swallow for both the groups.

As the results revealed that the clinical group had significantly higher scores than the control group in all the phases of swallow, the data was further analyzed to find the most severely affected phase of swallow in the clinical group. The feeding problems in children with Down syndrome were compared across the oral, pharyngeal and esophageal phase. The scores for each child were totaled for each subsection (oral, pharyngeal and esophageal phase) which was later converted to percentage scores since the number of questions in each subsection were different. The mean and standard deviation values for the clinical group were computed using descriptive statistics and have been depicted in table 4.2. The mean score for oral phase was highest followed by pharyngeal phase and esophageal phase.

The most commonly seen feeding problems in the oral phase in children with Down syndrome were absent jaw closure after receiving food, difficulty to form bolus and to control bolus, difficulty in moving the tongue laterally during bolus preparation, lack of awareness of food in mouth, retaining food in mouth without chewing, difficulty in chewing food, long duration for bolus manipulation and chewing, reduced anterior posterior tongue peristalsis, uncoordinated tongue movements and inappropriate oral transit time. Some of the most commonly seen problems in the pharyngeal phase in children with Down syndrome included retention of food in mouth after chewing without swallowing, pooling of food in lateral and anterior sulcus, aspiration/ choking during liquid intake and lack of awareness of residue in mouth. With respect to the esophageal phase some of the most commonly seen problems were burning sensation in mouth or throat after feeding and gagging sensation towards the end /after meals. Friedman test was conducted to determine whether there was a significant difference between scores

obtained in oral, pharyngeal and esophageal phase. Results of the analysis rendered a $\chi^2(2) = 5.29$, $p > 0.05$ which was not statistically significant. Hence, there was no evidence that the distribution of the three types of scores were significantly different. The mean values of the three phases of swallow for the clinical group have been depicted in figure 4.2.

Table 4.2.

Mean \pm standard deviation (SD) of the clinical group for the scores across phases of swallow.

Phases of Swallow	N	Mean \pm SD
Oral phase	17	20.92 \pm 15.77
Pharyngeal phase	17	14.14 \pm 10.15
Esophageal phase	17	10.78 \pm 17.61

Values are given as Mean \pm SD

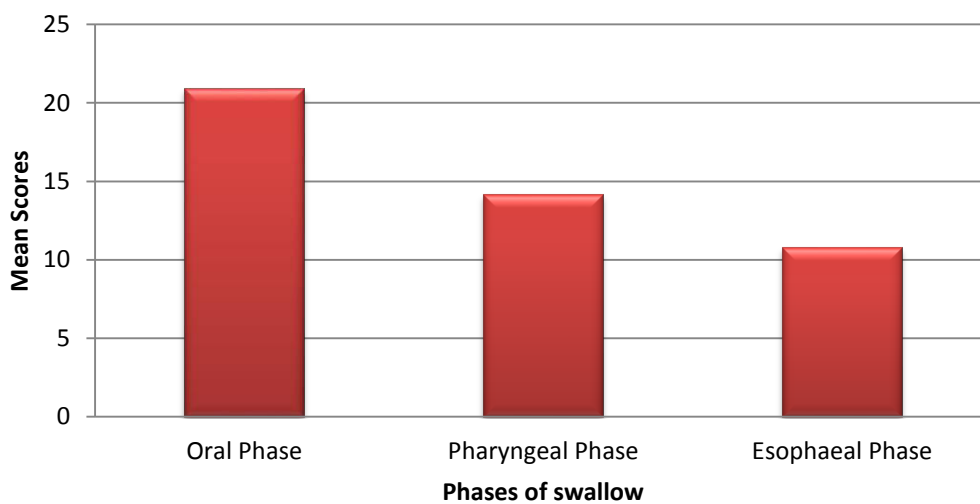


Figure 4.2. Mean scores across the phases of swallow in the clinical group.

Within the oral and the pharyngeal phases of swallow the feeding difficulties observed were scored separately for food items based on solids and liquids. The food items used were biscuit, banana and water. The mean score of solid food item (6.76 ± 4.75) was greater than liquid food item (3.58 ± 4.1) for the oral phase. For the pharyngeal phase the mean score of solid food item (4.76 ± 2.88) was again greater than liquid food item (1.17 ± 1.7). The overall total mean score was greater for solid than for liquid food item. A comparison of the scores for solid and liquid food items in the oral phase and pharyngeal phase were conducted using Wilcoxon Signed Ranks test. Results of this analysis indicated that scores obtained for solid versus liquid of the oral phase were significant ($z=3.31, p < 0.001$). Further the scores obtained for solid versus liquid of the pharyngeal phase also were significant, ($z=3.53, p < 0.001$).

The results of comparison between the two groups indicated that children with Down syndrome had significantly greater frequency of feeding difficulties. This result is in agreement with the studies done by Mohamed, Alhamdan, and Samarkandy (2013) and Mitchell, Call, and Kelly (2003) who also found feeding difficulties in children with Down syndrome. Hennequin, Allison, and Veyrune (2000) also found a higher frequency of oral health related problems in children with Down syndrome which impacted all of the oral functions during development. They found that suckling, swallowing, and chewing were delayed and the feeding pattern observed in children with Down syndrome was found to be an intermediate pattern between a primary suckle–swallow pattern and full rotary chewing. They found that these children chewed food for shorter periods due to lack of development of mature masticatory pattern. Frazier and Friedman in 1996 also found chewing and swallowing affected in children with Down syndrome.

In the present study the results revealed that the mean score for the oral phase was highest followed by pharyngeal phase and esophageal phase which indicated greater feeding difficulties in the oral phase followed by pharyngeal phase and esophageal phase in children with Down syndrome. In the oral phase 52.9% of children exhibited a developmentally immature chewing pattern in which the jaw moves up and down in a single plane (munching pattern). Among these children who exhibited munching pattern, 23.5 % exhibited lateral jaw movement while chewing and only 47.1% of the children exhibited mature rotary jaw movement. The oral manipulation for food items were difficult for these children leading to loss of food, poor bolus control and manipulation, and swallowing of large, poorly chewed morsels. Due to the delay in the initiation of oral movements and partly due to pauses within each masticatory cycle nearly 23.5 % of children with Down syndrome held food longer in the mouth without chewing. Swallowing was found to be compromised in children with Down syndrome due to lack of anterior seal and sweeping action of the tongue thereby making bolus transit inefficient. These children were adapted to the primitive forward-backward movement of the tongue against the palate to swallow food and as a consequence of the forward-backward movement, tongue thrusting while swallowing was evident in nearly 29.4% of the children with Down syndrome.

In the pharyngeal phase 29.4% of children retained food in mouth after chewing without swallowing. This could be attributed to the delay in the initiation of oropharyngeal movement of swallow. Three out of seventeen children demonstrated poor lip seal during swallowing leading to loss of food while chewing and swallowing. Few parents complained of aspiration (occasional) in their children for both solid and liquid

food item. The poor lateralization of tongue led to the pooling of food in the lateral and anterior sulcus in 64.7% of children with Down syndrome, who either removed it using a finger or by drinking some water. 52.9% of these children demonstrated lack of awareness of residual food in mouth after swallowing. 23.5% of children with Down syndrome indicated of food being stuck in lower throat and complained of burning sensation in mouth or throat.

A few problems with the esophageal phase were also seen in the clinical group. Only 11.7% occasionally vomited after feeding. This could be attributed to the swallowing of large poorly masticated morsels or partly due to the reduced muscle tone leading to poor digestion, esophageal obstruction and gastroesophageal reflux. The present result is in consensus with the studies conducted by Craig, Peter, and Joyce (1982); Bianca, Bianca, and Ettore (2002); Wallace, (2007) who had confirmed esophageal motor disorders such as abnormalities in esophageal peristalsis and in lower esophageal sphincter function leading to loss of food, food refusal, vomiting, gastroesophageal reflux disease, choking and weight loss in children with Down syndrome. The present study however does not show a statistically significant difference across the phases of swallow indicating that these children with Down syndrome faced feeding difficulties in all the three phases. In seventeen children with Down syndrome ten children had problems in the oral phase, eight children had problems in the pharyngeal phase, seven children had both oropharyngeal problems and only four had problems in the esophageal phase.

The results of comparison of solid food items versus liquid food items indicated that the children with Down syndrome faced greater feeding difficulties with solid food items than liquid food items in the oral and pharyngeal phases of swallow. In the present study the children with Down syndrome demonstrated delayed initiation of the sequence required for biting and chewing the semi-solid and solid textures. Hence the sequence of oropharyngeal functions necessary to move the semi-solid or solid food item from the lip region into the pharynx was poorly co-ordinated due to the impaired muscular control or co-ordination. The difficulty in biting and chewing solids adequately could be, partly because of incoordinated tongue movements and partly because of poor control. In the present study nearly 41% of children with Down syndrome demonstrated difficulty in biting while 47% demonstrated difficulty in chewing solids and semisolids. Due to absent lip seal and poor coordination and control of jaw movements these children demonstrated loss of water, poor bolus control and manipulation on drinking liquids. The transitions of jaw movements in these children were also found to be slow.

Nine out of seventeen children had a previous history of feeding difficulties where they had difficulties with cup drinking, straw drinking, advancing food textures and difficulty in chewing and biting semi-solid and solid food items. This could be attributed to the limited the range and coordination of jaw and tongue movements which affected the gain of efficient chewing skills. Instead of the distinct lateral shifting of bolus from midline on to the molar surfaces and again back to the midline these children with Down syndrome preferred to have a quick and rolling tongue movement to lateralize food. Only a small percentage of children were able to precisely move solid food from side to side.

The present study is in consensus with the study conducted by Mohamed, Alhamdan, and Samarkandy (2013) who found that the children with Down syndrome had difficulties in transition from liquids to solids. Gisel, Lange, and Niman (1984) revealed that children with Down syndrome exhibited a reluctance to chew and preferred sucking of food and decreased masticatory efficiency when it comes to chewing solids. They found that children with Down syndrome due to difficulties in chewing solids tend to swallow their food which may lead to overeating in order to compensate for the loss of gustatory input. In the present study too children with Down syndrome had a greater difficulty in chewing and biting solids. The feeding problems in children with Down syndrome could be due to the inadequate jaw movements, inadequate lip closure, restricted tongue movements, inadequate biting and chewing, difficulty in forming and controlling the bolus, difficulty in moving the tongue laterally during the bolus preparation, restricted anterior-posterior tongue peristalsis and uncoordinated tongue movements which was more predominantly affected for solids than liquids.

The feeding difficulties exhibited by the children with Down syndrome could be attributed to the oromotor skills. The children with Down syndrome can demonstrate both sophisticated and unsophisticated oromotor patterns as they progress in the development of oromotor skills. In the present study Com-DEALL Oro Motor Checklist (Archana & Karanth, 2008) was administered to assess the oro-motor skills in both the groups. Three domains i.e. jaw movement, tongue movements and lip movement with a total of 24 questions were considered for evaluation. Responses were rated on a three point rating scale where '0' signified absent, '1' signified only spontaneously present and '2' signified consistently present. A higher score on this checklist indicates better oro-motor

abilities. The scores obtained for each child for each domain in both groups was totaled. The mean and standard deviation values for the clinical group and the control group were computed using descriptive statistics and have been depicted in table 4.3. On comparison, it was seen that the mean scores for oro-motor abilities were greater for the control group than for the clinical group indicating greater oromotor problems in clinical group. To check whether a significant difference existed between the two groups, Mann-Whitney U test was used. The results revealed that the clinical group had significantly higher scores than the control group. The |z| values have been depicted in table 4.3.

Table 4.3

Mean±standard deviation (SD) and |z| value of both the groups for the scores across oromotor abilities and phases of swallow.

Oromotor skills	N	Clinical Group	N	Control Group	 z value
Jaw movements	17	8.70±2.86	47	11.89±0.42	5.55*
Tongue movements	17	12.70±3.85	47	19.36±1.46	6.33*
Lips movements	17	13.35±3.04	47	15.57±0.94	3.52*
Total Score	17	34.76±8.32	47	46.82±2.52	6.15*

Values are given as Mean±SD, * p < 0.001

Since the clinical group demonstrated oro-motor problems the data was further analyzed to see which structure was most affected and thereby could have contributed to the feeding and swallowing difficulties. The oro-motor problems in the clinical group were compared across the jaw, tongue and lip movement. The mean and standard deviation values for the clinical group were computed using descriptive statistics and

have been depicted in table 4.4. The mean value for lip movement was highest followed by jaw movement and tongue movement. Friedman test was conducted to determine whether there was a difference in scores obtained across the jaw, tongue and lip movement. Results of the analysis rendered a $\chi^2 (2) = 7.614$, $p < 0.05$ which was statistically significant. A post hoc comparison of the scores of the oro-motor abilities was conducted using Wilcoxon Signed Ranks test. Results of this analysis indicated that the score for tongue movements versus lip movement was significant ($z=3.15$, $p < 0.05$), and the score obtained on lip movement versus scores of jaw movement was also significant ($z=2.04$, $p < 0.05$). However the scores obtained on lip movements versus the scores of tongue movement was not significant ($z=1.22$, $p > 0.05$). The mean scores obtained across the three domains for the clinical group has been depicted in figure 4.3.

Table 4.4

Mean±standard deviation (SD) of the clinical group for the scores across oromotor abilities.

Oro-motor skills	N	Mean ±SD
Jaw movement	17	72.54±23.89
Tongue movement	17	65.00±21.06
Lip movement	17	83.45±19.00

Values are given as Mean±SD

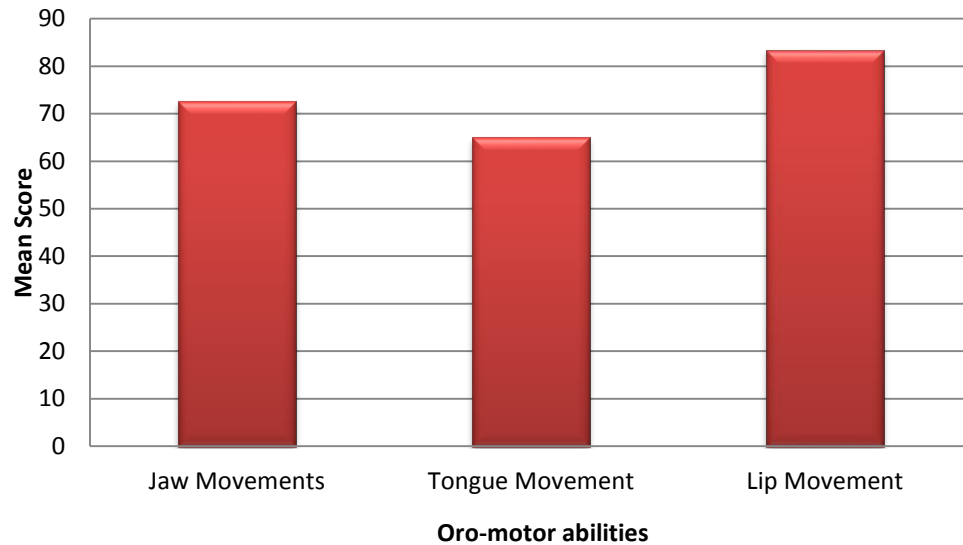


Figure 4.3. Mean scores across the three domains in the clinical group.

The results of Com-DEALL Oro Motor Checklist across both the groups revealed that oromotor problems were significantly higher in the clinical group. In the present study, in each domain the following were predominantly observed in children with Down syndrome. In domain of jaw movements, difficulties in biting and chewing solid food, clenching of teeth, open mouth posture and reduced transition from one movement to another were the main problems faced by children with Down syndrome. In the domain of tongue movements, restricted lateralization towards left and right of the mouth (outside and inside) and reduced tongue tip elevation was observed. With regard to the lip movements, these children with Down syndrome mainly faced difficulties in chewing with lips closed, sucking through straw and blowing with a proper lip rounding. The open mouth posture seen in 18% of the children with Down syndrome in the present study during rest could be attributed to the low muscle tone in the lips, and tongue, jaw instability or loose ligaments in the temporomandibular joint. The results indicate that children with Down syndrome faced problems in oro-motor abilities which in turn could have impacted the feeding, eating and drinking.

The result supported many of the anatomical and physiological findings that have been reported in literature regarding the associated characteristics of children with Down syndrome. Fischer-Brandies (1989) and Stevenson and Allaire (1991) also promulgated that the normal development of oral structure and its function is altered in infants and children with Down syndrome which in turn leads to the development of a compromised suckling, swallowing, and mastication. Arvedson and Brodsky (2002) also reported that the feeding problems could be consequent to the abnormal oral muscle tone and strength. Rogers, Arvedson, Buck, Smart, and Msall (1994) also attributed the feeding problems to restricted or-motor abilities. The present study is also in consensus with studies conducted by Mitchell, Call, and Kelly (2003); Field, Garland, and Williams (2003); where they discovered a significantly higher prevalence of oral-motor problems in children with Down syndrome than in the other children without Down syndrome.

The results of Com-DEALL Oro Motor Checklist across each domain (jaw, tongue and lip movement) revealed that the tongue movements were highly affected followed by jaw movement and lip movement. In the present study nearly 58% of children with Down syndrome had difficulty in lateralizing the tongue outside the mouth and 82% of them had difficulty in lateralizing the tongue inside the mouth. Tongue tip elevation was affected in 65% of the children with Down syndrome indicating the impairment in muscle control and coordination which led to restricted tongue movements, poor tongue coordination, and decreased jaw strength making it difficult to chew and bite on solids.

The poor oro-motor abilities could also be associated with oral hypotonicity leading to restricted tongue movements. In the present study, eleven children with Down syndrome were diagnosed to have hypotonicity. Hypotonicity can lead to postural instability resulting in insufficient head, neck and trunk control and can also directly affect eating and drinking. In the present study hypotonicity in the lips was demonstrated in 47% of the children with Down syndrome, while hypotonicity in the tongue was exhibited in 57% of the children with Down syndrome. The large hypotonic tongue present in 7 children with Down syndrome coupled with the poor oro-motor skills reduced the ability of the tongue to crush food against the palate, lateralize food towards the molar and back to the blade of the tongue and form a cohesive bolus. This may lead to the swallowing of large inadequately masticated food.

In the present study the reports based on physiotherapy evaluation, feeding history and results of Com-DEALL Oro Motor Checklist revealed that hypotonicity seen in these children has contributed to the open mouth posture; difficulty in jaw closure and subsequent reduced lip closure; difficulty in forming tight lip seal for sucking; difficulty with precise tongue movements; difficulty with dynamic jaw and tongue stability as well as in fine graded jaw, tongue and lip movements which impact eating and drinking. This is in consensus with the study conducted by Kumin and Bahr (1999) who revealed different degrees of hypotonia in different oral structures. They found in their study that 44% children with Down syndrome demonstrated low muscle tone in the lips, while low muscle tone in the tongue was exhibited in 80% of the children. The open mouth posture seen in 18% of the children with Down syndrome in the present study during rest could be related to the low muscle tone in the lips, jaw instability or loose ligaments in the

temporomandibular joint. The present study is also in consensus with the study of Shapiro et al., (1976) who reported that the lingual hypotonicity in children with Down syndrome presents as an obstacle to the development of oral–motor coordination thereby leading to compromised feeding.

Sensory issues can also affect mastication leading to poor oral exploration, difficulty in transitioning to food of different textures and an inability to initiate, grade, or sustain oral patterns for feeding and swallowing. In the present study, 35.3% of children with Down syndrome demonstrated difficulty in transitioning to varied textured food. These children had a cluster of behaviors associated with oral hyposensitivity such as poor or lack of awareness of food on lips and tongue, pocketing of food in the anterior and lateral sulcus, stuffing of food in the mouth and retention of food in mouth without chewing. 17% of children with Down syndrome demonstrated reduced perception of light touch on the gum. Thus the feeding difficulties in children with Down syndrome could also be attributed to sensory issues. Frazier and Friedman in 1996 study also revealed that children with Down syndrome exhibited poor or no awareness of food on lips, slow registration of food in mouth, pocketing of food and stuffing of mouth indicating the cluster of behaviors associated with oral hyposensitivity. The result of the present study also agrees with the finding by Field, Garland, and Williams (2003) who found a significantly higher prevalence of oral-motor problems, swallowing difficulties, and texture selectivity in children with Down syndrome, which suggested that they had the ability to chew but refused to do so as a result of learned aversions to specific textures that prompted unpleasant experiences such as gagging and vomiting.

In addition, the typical craniofacial characteristics present in the clinical group such as class III malocclusion, tongue thrust, large tongue size and a reduced number of teeth could have further decreased the masticatory capacity. The combination of craniofacial features and reduced muscle tone has led to compromised feeding and swallowing in the clinical group. Malocclusion which was present in 8 children with Down syndrome could have significantly impaired chewing and grinding of food thereby impacting eating, drinking and swallowing process. Other factors that impaired feeding could be frequent ulcers, plaque and debris accumulation and poor oral hygiene which increase the possibility of caries rate as well as gradually cause acute infection, inflammation, and pain in the periodontal areas resulting in absolute refusal of food or the tendency to swallow food whole.

The feeding problems were rated for its severity in each phase by the clinician on a 5 point rating scale. The severity rating varied from 0-4 where, '0' represented no difficulty at all; '1' represented mild feeding difficulty; '2' represented moderate feeding difficulty; '3' represented moderately severe feeding difficulty; and '4' represented severe feeding difficulty. Based on the rating of the oral phase it was seen that out of seventeen children ten were rated to have mild degree of feeding problems, four were rated to have moderate degree of feeding problems and six were rated to have no feeding difficulties. In the pharyngeal phase out of seventeen children seven were rated to have mild degree of feeding problems, two were rated to have moderate degree of feeding problems and five were rated to have no feeding difficulties. In the esophageal phase out of seventeen children three were rated to have mild degree of feeding problems, one was

rated to have moderate degree of feeding problems and twelve were rated to have no feeding difficulties.

The overall severity of feeding and swallowing difficulties was assessed at the end of the section four of the questionnaire, where the parents/caregivers (main feeders) were asked to rate the overall severity of their child's feeding and swallowing for eating and drinking on a 5 point rating scale. The overall severity of feeding problems faced by the child with Down syndrome was also rated by the investigator. On comparison of the investigator and caregiver rating it was seen that only one out of seventeen parents of children with Down syndrome had rated the child to have moderate degree of feeding difficulties, whereas sixteen parents rated the child to have no problems in feeding and swallowing for eating and drinking. However the clinician rated two children to have moderate degree of problems in feeding and swallowing for eating and drinking, nine children to have mild degree of problems in feeding and swallowing for eating and drinking and six children to have no problems in feeding and swallowing for eating and drinking. This indicated the parent's lack of awareness of feeding difficulties faced by their children. Though these children with Down syndrome faced feeding difficulties, none of the parents made any kind of modification to the utensils used for feeding and positioning and seating during feeding. Only three out of seventeen parents made an alteration to food temperature and only seven caregivers restricted the quantity of food per mouth to suit the child's needs. The present study is in agreement with the study conducted by Spender, Stein, Dennis, Reilly, Percy, and Cave, 1996 who found in their routine clinical interviews that none of the parents of children with Down syndrome had mentioned that their children had eating difficulties indicating that parents either have

simply adapted to the feeding problems or developed strategies that ensured adequate nutritional input. Consequently the feeding problems are generally overlooked and neglected. Hence the complexities and issues related to feeding difficulties in infants and children with Down syndrome should be addressed since birth onwards by a team of professionals such as speech-language pathologist, physio/occupational therapist, psychologist, nutritionist, and pediatrician who have specialized knowledge, skills and sensitivity to the identification and intervention of feeding difficulties. Family members and caretakers should be trained to be sensitive to the feeding difficulties in their own infants and children with Down syndrome. It is important to educate the parents/caregivers (main feeders) on understanding the child's nutritional needs, importance of acquiring a sense of competence in meeting a healthy lifestyle, including healthy feeding behaviors and to promote short term and long term health of their child.

III. Comparison of feeding abilities between age groups of children in both the groups

The clinical group and the control group were divided into two age groups (2-5 years and 5.1-7 years). In the clinical group eight children were grouped into the lower age group (2-5 years) and nine children were grouped into the higher age group. In the control group thirty children were grouped into the lower age group (2-5 years) and seventeen children were grouped into the higher age group. The mean and standard deviation values for both the two age groups (2.5 and 5.1-7 years) in the clinical and control groups were computed using descriptive statistics and have been depicted in table 4.5. and table 4.6. The scores obtained across the phases of swallow were compared in

both the age groups. In the clinical group higher mean scores were obtained by the lower age group (2-5 years) in comparison to the higher age group (5.1 -7 years). Mann-Whitney U test revealed that there was no significant difference between the two age groups for the scores obtained across the phases of swallow. In the control group too higher mean scores were obtained by the lower age group (2-5 years) in comparison to the higher age group (5.1 -7 years). Mann-Whitney U test showed that there was a significant difference between the two age groups for the scores obtained across the oral phases (solids) and pharyngeal phase (solids) of swallow. The |z| values for the two age groups of the clinical group have also been shown in table 4.5. and table 4.6. The performance of both the groups in the clinical group and the control group have been depicted in figure 4.4. and figure 4.5

Table 4.5.

Mean±standard deviation (SD) and |z| value of both the age groups in the clinical group for the phases of swallow.

Phases of swallow	N	2-5 years	N	5.1-7 years	 z value
Oral phase (Solids)	8	8.00±3.70	9	5.66±5.50	0.58*
Oral phase (Liquids)	8	3.75±0.71	9	3.44±5.27	0.89*
Pharyngeal phase (Solids)	8	4.50±1.92	9	0.88±1.53	0*
Pharyngeal phase (Liquids)	8	1.5±1.92	9	0.88±1.53	1.23*
Esophageal phase	8	0.75±1.03	9	0.55±1.13	0.48*

Values are given as Mean±SD, * p >0.05

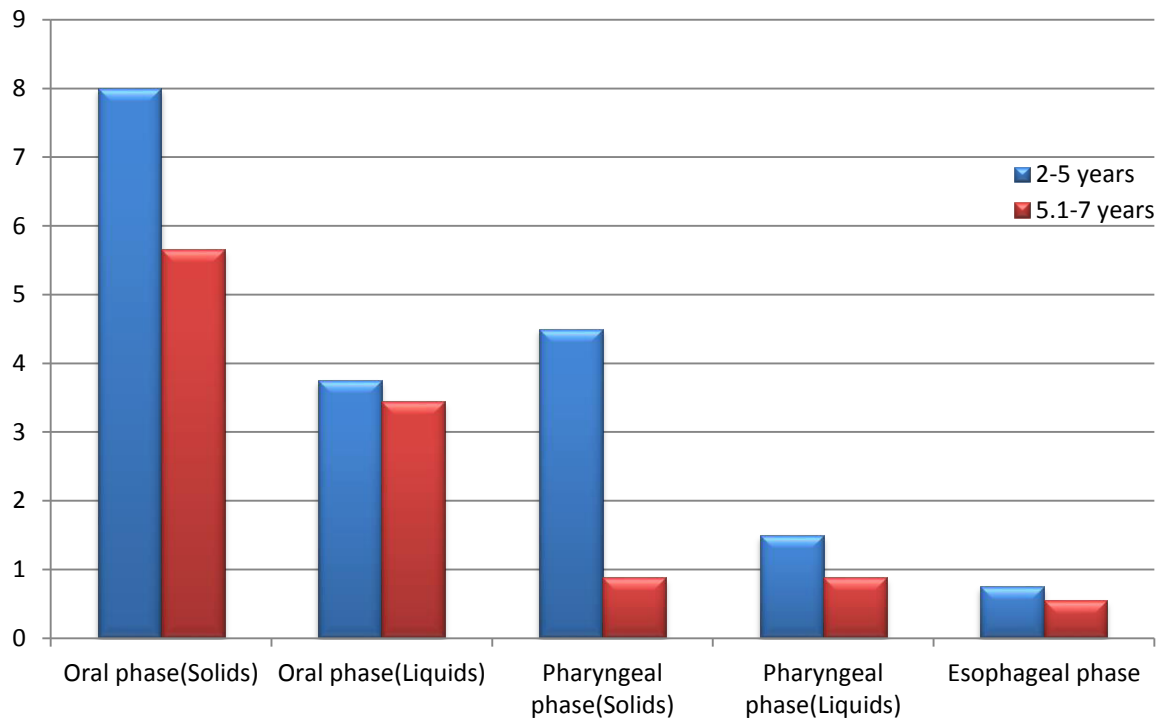


Figure 4.4. Mean scores of both age groups across the phases of swallow in the clinical group.

Table 4.6.

Mean±standard deviation (SD) and |z| value of both the age groups in the control group for the phases of swallow.

Phases of swallow	N	2-5 years	N	5.1-7 years	 z value
Oral phase (Solids)	30	0.53±0.89	17	0	2.47*
Oral phase (Liquids)	30	0.06±0.25	17	0.05±0.24	0.10**
Pharyngeal phase (Solids)	30	0.53±0.97	17	0	2.63*
Pharyngeal phase (Liquids)	30	0.06±0.25	17	0	1.07**
Esophageal phase	30	0.06±0.36	17	0.11±0.48	0.41**

Values are given as Mean±SD, * p < 0.05, **p > 0.05

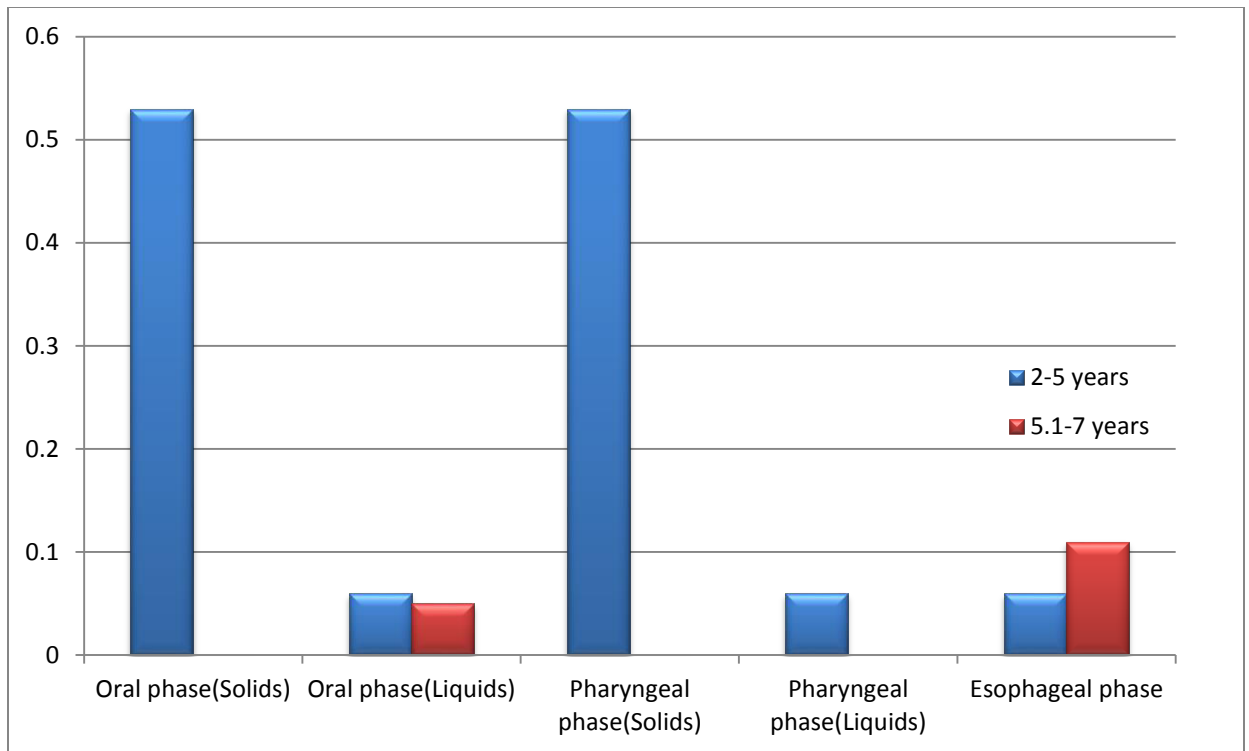


Figure 4.5. Mean scores of both age groups across the phases of swallow in the control group.

The results revealed that in the clinical group higher mean scores were obtained by the lower age group (2-5 years) in comparison to the higher age group(5.1 -7 years) indicating that feeding difficulties were present to a greater extent in younger children (2-5 years) than the older children with Down syndrome. This could be attributed to the following factors. First, the masticatory pattern observed in younger children with Down syndrome was a transitory pattern from munching to more matured rotary chewing. Due to the pre dominance of munching pattern of chewing, mastication was inefficient leading to the poor bolus formation and control of cohesive bolus. Secondly it was observed that younger children with Down syndrome demonstrated significantly higher prevalence of oral-motor problems, food refusal, narrow food preferences, texture selectivity and

swallowing difficulties. The oral-motor problems seen in the younger children with Down Syndrome included poor coordination and sequencing of oro-motor movements, delayed initiation of tongue and jaw movements, difficulty grading jaw movements for mastication, restricted range of tongue movements such as lateralization and elevation. Thirdly, these problems in particular were more prevalent in those children with craniofacial anomalies, cardiac or pulmonary conditions. The marked craniofacial dysmorphology in younger children with Down syndrome significantly delays the maturation of mastication coordination and chewing efficiency. The craniofacial features such as macroglossia interfered with adequate movements of the tongue within the small size of the oral cavity. The positioning of the characteristic hypoplastic maxilla and protruding mandible can effectively lock the jaw in place and prevent lateral movements for chewing. Few of the parents/ caregivers of children with Down syndrome preferred giving mashed food or semisolid food items due the presence of cardiac issues in these children. Respiratory issues seen in these children resulted in more pronounced tongue protrusion patterns that interfered with eating solids. This can also compromise with the maturation of advanced tongue patterns to support chewing. Hence these children either refused solids or preferred food items that they can easily swallow. The feeding history revealed that younger children were more prone to aspiration leading to complications such as respiratory illness, congestion and pneumonia. Fourthly, the hypotonicity of the muscles led to delayed initiation, poor coordination and sequencing of oromotor movements, difficulty grading jaw movements for chewing, weak lip closure, and weak and reduced tongue movements. Finally feeding difficulties were more predominant in

younger children with Down syndrome as the influence of intervention was not evident as they only received it for a shorter duration.

Children in the older age range demonstrated fewer feeding problems which could be attributed to the following factors. First, in the older group the pattern of chewing is more matured and moving towards the more rotary pattern which could have brought about the reduction in feeding problems. Moreover as they mature, probably the coordination between oral structures could have improved and the individual movement of the articulatory structure could have improved. Secondly the children in the higher age group had fewer oro-motor problems, wider food preferences and no texture selectivity. As the oromotor coordination and sequencing of movements improve with age, these children demonstrated fewer problems with initiation, grading and sequencing of movements required for feeding, chewing and swallowing. Thirdly, as these children grow, the increased bruxism seen in them, help them to cope up with the problems associated with malocclusion, thereby reducing the difficulties in mastication (Macho et al., 2008). Lastly, the influence of intervention could have reduced the marked hypotonicity in these children thereby improving the coordination and control of oral structures during feeding.

The present study is in consensus with the studies conducted by Hennequin, Allison, and Veyrune (2000); Kumin, (1994); Pipes, (1995); Cooper-Brown et al., (2008) who found greater prevalence of feeding difficulties in younger children because of the functional impairment which impacted all the oral functions during maturation such as suckling, swallowing, and chewing abilities. In the present study five out of nine children with Down syndrome in the older age range (5.1-7 years) had a previous history of

feeding difficulties such as difficulty in transitioning from breast/bottle feeding to cup feeding, and from liquids to semisolids to solids, difficulties in chewing and biting solid food, difficulty in swallowing food and aspiration. Out of five children two of the children with Down syndrome still had persisting feeding difficulties indicating the importance of early assessment and intervention of the feeding difficulties in children with Down syndrome.

In the control group too the higher mean scores were obtained by the lower age group (2-5 years) in comparison to the higher age group (5.1 -7 years). Various studies have shown that only a small percentage of children over 2-5 years precisely move solid food from side to side, instead they tend to have a preference on slow and rolling tongue movements to lateralize food (Gisel, 1988). As the child grows older the lateral movements of the tongue is established and the complexity of jaw movements increases which helps in transferring the bolus to the molar or chewing surfaces. Vitti and Basamajia, (1975) reported that normally the maturation of mastication coordination in typically developing children is fully achieved by 6 years of age. Gisel and Patrick (1988) also reported that the time taken for chewing the solid food gets lesser as the child grows older. Children refine their oral skills, expand the different texture of foods they accept, become more precise and efficient at chewing food that require more extensive oral manipulation and also start handling liquids from open cups from 12-36 months of age. Further the refinement of independent self- feeding skills occurs only after 2 years of age (Delaney & Arvedson, 2008).

IV. Comparison of feeding difficulties across gender in both the groups.

The mean and standard deviation values for both the gender in both the age ranges in both the groups were computed using descriptive statistics. The mean and standard deviation obtained for both the groups have been depicted in table 4.7. and table 4.8. On comparison, it was seen that the mean scores of all phases of swallow (oral, pharyngeal and esophageal phases) were higher for the males than females of the clinical group in the higher age range (5.1-7 years) whereas the mean scores for the lower age range (2-5.1years) varied only marginally across gender in the clinical group. On comparing the clinical group as a whole across gender the mean values were greater for males than for females, indicating greater feeding difficulties in males than females.

In the control group too, it was seen that the mean scores of all phases of swallow (oral, pharyngeal and esophageal phases) were higher for the males than for females in the lower age range (2-5.1years) whereas the mean scores for the higher age (5.1-7 years) range varied marginally across gender in the control group. On comparing the control group as a whole across gender the mean values were greater for males than for females, indicating that males exhibit greater feeding difficulties when compared to females. To check whether a significant difference existed between the two groups, Mann-Whitney U test was used. The results revealed no significant difference across gender in both the age groups in both the groups. The |z| values have been depicted in table 4.7. and 4.8.

Table 4.7.

Mean±standard deviation (SD) and |z| value for both gender in both age groups for the phases of swallow in the clinical group.

Phases of swallow	2-5 years				5.1-7 years				z value
	N	Male	N	Female	N	Male	N	Female	
Oral phase (Solids)	4	8.25±2.06	4	7.75±5.25	5	7.60±6.22	4	3.25±3.86	1.63*
Oral phase (Liquids)	4	3.50±2.88	4	4±2.94	5	6.20±5.84	4	0	2.07*
Pharyngeal phase (Solids)	4	4.75±2.50	4	4.25±1.5	5	6.40±4.03	4	3.25±2.50	0.95*
Pharyngeal phase (Liquids)	4	2±2.7	4	1±0.81	5	1.60±1.81	4	0	2.30*
Esophageal phase	4	0.5±1	4	1±1.15	5	1.00±1.41	4	0	1.75*

Values are given as Mean±SD, * p > 0.05

Table 4.8.

Mean±standard deviation (SD) and |z| value of both gender in both age groups for the phases of swallow in the control group.

Phases of swallow	2-5 years				5.1-7 years				z value
	N	Male	N	Female	N	Male	N	Female	
Oral phase (Solids)	17	0.41±0.71	13	0.69±1.10	10	0	7	0	0.34*
Oral phase (Liquids)	17	0	13	0.15±3.75	10	0.10±0.31	7	0.05±0.24	0.86*
Pharyngea l phase (Solids)	17	1.22±0	13	0.38±0.50	5	0	7	0	0.30*
Pharyngea l phase (Liquids)	17	0.24±0	13	0.07±0.27	5	0	7	0	0.21*
Esophagea l phase	17	0.48±0	13	0	5	0.20±0.63	7	0.11±0.48	1.23*

Values are given as Mean±SD, * p > 0.05

The present study does not statistically show a significant difference across gender in both the age groups in both the clinical and control group. The results of the clinical group hence suggest that feeding difficulties are equivalently present in both the age ranges irrespective of gender. In the lower age range (2-5 years) in the clinical group the mean values obtained across both the gender varied marginally indicating the

presence of feeding difficulties in both males and females. However in the higher age range the mean values obtained for males were higher than females indicating lesser feeding difficulties in females than males in the higher age range. The overall mean values were greater for males when compared to females in the clinical group indicating that males had more feeding difficulties compared to females.

In the control group too the overall mean values were greater for males when compared to females. However because of the lack of evidence in literature, it is currently difficult to speculate the influence of gender on feeding function in children with Down syndrome.

V. Influence of intervention in the clinical group

The intervention received by the children with Down syndrome was mainly speech and language therapy, physiotherapy, occupational therapy and special education. The influence of intervention for a duration of less than 2 years (n=10) and greater than 2 years (n=7) was compared within the clinical group for the total scores of phases of swallow obtained on the questionnaire. The mean and standard deviation obtained have been depicted in table 4.9. On comparison, it was seen that the mean scores (scores for oral, pharyngeal and esophageal phases) were higher for the clinical group who had attended intervention for less than 2 years than for those who had attended for greater than 2 years. However the results on Mann-Whitney U test revealed no statistically significant difference in the clinical group. The $|z|$ values have been depicted in table 4.9. The mean of the scores obtained across the phases of swallow for different duration of intervention have been depicted in figure 4.6.

Table 4.9.

Mean+standard deviation (SD) and |z| value of scores based on the duration of intervention in the clinical group.

Phases of swallow	Intervention <2 years	Intervention >2 years	z value
Oral phase (Solids)	8.60±3.23	4.14±5.55	1.61*
Oral phase (Liquids)	4.20±3.82	2.71±4.71	1.21*
Pharyngeal phase (Solids)	5.60±2.71	3.57±2.87	1.68*
Pharyngeal phase (Liquids)	1.40±1.89	0.85±1.46	0.73*
Esophageal phase	0.70±1.15	0.57±0.97	0.18*

Values are given as Mean±SD, * p > 0.05

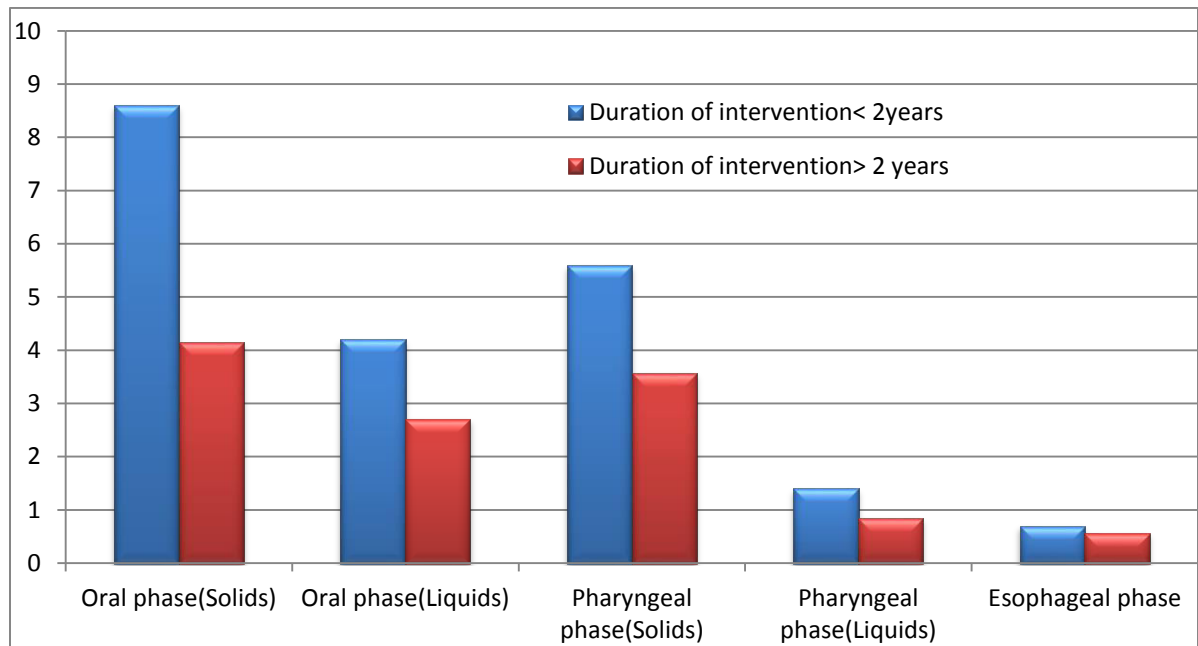


Figure 4.6. Mean scores based on the duration of intervention across the phases of swallow in the clinical group.

The intervention received by the children with Down syndrome was mainly speech and language therapy, physiotherapy, occupational therapy and special education. Amongst those children who had received intervention for less than 2 years (n=10), few children had attended speech and language therapy, physiotherapy, occupational therapy and preschool special education. Whereas in those children who had received intervention for more than 2 years (n=7), most of them had attended speech and language therapy, physiotherapy, occupational therapy and preschool special education. In those children who had received intervention for more than 2 years, a greater number of children had attended intervention which could have impacted on overall growth and development of the child. This could in turn have influenced on feeding development as well, thereby resulting in lower scores in the phases of swallow in comparison with those children who had attended intervention for less than 2 years. Besides it could also indicate the persistence of feeding problem in those who had received intervention for less than 2 years indicating the need to include feeding therapy in the treatment protocol. However the present study does not statistically show a significant difference indicating that in the intervention feeding aspects were not focused. Since the caregivers were not aware of the child's feeding difficulties which was evident from overall severity rating, they did not even report of the feeding problems to the clinician and therefore no specific strategies were taken up during intervention to tackle feeding issues. Literature addressing the influence of intervention is scarce; hence there is a dearth of literature examining the effects of intervention on feeding function in children with Down syndrome. Hence in order to provide effective early communication intervention services from birth, the knowledge, skills and sensitivity of speech language therapists to identify

caregiver needs, to provide appropriate family-focused intervention and to make recommendations regarding the management of feeding problems in infants and children with Down syndrome is of great importance.

In sum, the results of the present study indicated that feeding problems were significantly higher in the oral, pharyngeal and esophageal phases for the clinical group in comparison to the control group. The feeding problems in children with Down syndrome were highest in the oral phase followed by pharyngeal and esophageal phase. Within the oral and the pharyngeal phases of swallow the feeding difficulties were significantly greater for solids than liquids. It was also found that parent's/caregivers lacked awareness of feeding difficulties faced by their children. The feeding problems in the clinical group could be attributed to the oromotor problems which were present to a significantly greater extent than the control group. Further, the tongue movement was affected to a greater extent compared to the jaw and the lip movement in the children with Down syndrome. Age wise comparison in the clinical and control group indicated higher mean scores in the lower age group (2-5 years) in comparison to the higher age group (5.1 -7 years). However no significant difference was seen. Further the duration of intervention did not influence the feeding abilities.

Chapter 5

Summary and Conclusion

Feeding which is a crucial process for the overall growth and development of the child, is frequently found to be affected in children with Down syndrome. These deficits could arise due to the structural abnormalities which affect the normal development of oral structures and its functions in infants and children with Down syndrome which in turn leads to the development of a compromised suckling, swallowing, biting and chewing. Studies have revealed a variety of problems faced by infants and children with Down syndrome such as problems with breast and bottle feeding; a poor or delayed suckling ability, problems with mastication, drooling, slow development of the ability to manipulate food in fingers and in the use of feeding utensils; the failure to progress through a normal sequence of food textures and the refusal of certain foods, particularly those of a hard texture; behavioral problems such as the refusal to swallow, the spitting out of food, or the retention of food or utensils in mouth. Although some western studies have been carried out to identify the nature of feeding problems in children with Down syndrome, these are limited. Most of these studies are on infants and focus on the breastfeeding difficulties and oromotor deficits in them. A very few studies objectively assess feeding difficulties in the oral and pharyngeal phases. There are much lesser studies which objectively assess all the three phases of swallow to assess feeding related issues. Hence this study would add to the objective evaluation of feeding difficulties in all the three phases of swallow in children with Down syndrome. Further, as mentioned above, most of the studies have been carried out on infants with Down syndrome. Since

feeding is a skill that develops by 2 years of age and refines till 6 years of age (Delaney & Arvedson, 2008), it is essential to study the children in this age group as well.

The paucity of literature makes it clear that there are deeper underlying complex issues pertaining to feeding in children with Down syndrome that needs to be investigated. A more in depth study covering various aspects related to feeding is to be conducted. No study has undertaken the in-depth quantitative assessment of all the three phases of swallowing. A more effective and practical assessment of a child's ability to feed would account for the efficient management and reasonable adjustments to be made to know the quality of life. Therefore, the present study was undertaken with the aim of assessing the feeding problems, if any, in children with Down syndrome in the age range of 2-7 years. The specific objectives of this study were to compare the feeding problems in children with Down syndrome with that of the typically developing children, to compare the feeding abilities between the lower vs. higher age group of children in both groups and to compare the feeding difficulties across gender in both the groups. The study also investigated the influence of intervention on feeding problems in children with Down syndrome.

The study was carried out in three phases. The initial phase involved the development of a questionnaire to assess feeding problems faced by children with Down syndrome. This was prepared by collating information from the literature and based on the complaints concerning feeding received from the clients registered in the Special clinic for motor speech disorders, Department of Clinical Services, All India Institute of Speech and Hearing, Mysuru. The questions focused on the physical problems faced by the children during feeding and swallowing. There were questions which focused on

orosensorimotor issues, general feeding issues, feeding history, modifications during feeding and the different phases of swallow. The questions were grouped under four sections: I. Demographic data and general history, II. Craniofacial and orosensory assessment, III. Feeding history and IV. Assessment of different phases of swallow.

The content validity of the questionnaire and the rating scale was assessed by obtaining the feedback from three experienced speech-language pathologists. They were asked to judge the appropriateness of the questions included and the rating scale used. A pilot study was carried out on 10 children with Down syndrome and on 10 typically developing children. The questionnaire was administered on two parents/caregivers of children with and without Down syndrome in different age groups between the age range of 2-7 years. Following this the final version of the questionnaire was prepared after the content validation.

The final version of the questionnaire was administered on 17 children with Down syndrome (8 females and 7 males) and 47 typically developing children (20 females and 27 males) in the age range of 2-7 years. The children with Down syndrome were amongst those who reported to the Department of Clinical Services, All India Institute of Speech and Hearing, Mysuru, participated in the study. They were diagnosed as 'Delayed Speech and language with Down Syndrome' by a qualified team of professionals including speech-language pathologist, pediatrician and a clinical psychologist. The degree of Intelligence quotient ranged from near normal intelligence to moderate retardation. These participants were further subdivided into two groups based on their age. There were 9 children in the higher age group (5-7years) and 8 in the lower age group (2-5 years). They constituted the clinical group. A group of forty seven typically developing children

(20 females and 27 males) matched for age and socioeconomic status were selected. There were 17 children in higher age group (7 females and 10 males) and 30 in the lower age group (13 females and 17 males).

The testing was carried out in a relatively noise free environment with minimum distractions. Each child was tested individually. A rapport was established with the mother/caregiver. The purpose of the administration was explained. The demographic data was obtained initially. The WHO Ten-question disability screening checklist was administered on typically developing children to rule out history of neurological, oromotor, communicative, cognitive, or sensorimotor and academic impairment. The Assessment Checklist for speech-language domain was also administered on the typically developing children to assess age adequate language abilities. The oro-motor abilities was assessed by administering the Com-DEALL Checklist for Assessment of Oro-motor skills in Toddlers (Archana & Karanth, 2008) which was followed by the administration of the final version of the questionnaire developed. A video recording of the child's feeding skill was carried out in order to facilitate a better view of the feeding problems presented by the children. The child was given different items to eat and drink (e.g., Biscuit, banana, water etc.) to permit a first-hand observation of the feeding skills.

Each statement in the IInd, IIIrd and IVth section was accompanied with a response choice of “no” (a score of zero) or “yes” (a score of 1). A 5 point rating scale was also prepared to assess the severity of the feeding problems in the IVth section of the questionnaire at the end of every phase of swallow. The feeding problems faced by the child were noted. At the completion of the section four of the questionnaire the parents/caregivers were asked to rate the overall severity of their child's feeding and

swallowing for eating and drinking separately on the 5 point rating scale. The time taken to administer the questionnaire was approximately 45 minutes. On the whole to administer all the necessary protocols, the time taken was approximately 60 minutes. Positive reinforcements like verbal and social reinforcements were provided to maintain the interest and motivation of the child throughout the test administration.

The total scores obtained in section IV and the sub section scores (oral, pharyngeal and esophageal phase of swallow) from all the participants were fed to the computer for statistical analysis. SPSS version 20 software was used for the statistical analysis. Descriptive statistics was used to obtain mean, median and standard deviation of scores obtained on the questionnaire for both the groups. Shapiro–Wilk test was administered to check for normality. Since the data did not follow a normal distribution and due to high standard deviation, nonparametric tests were used for statistical analysis. Mann-Whitney U test was used to compare between the clinical group and control group and to measure the influence of different independent variables. Friedman’s test and Wilcoxon Signed Rank Test was used to measure the influence of different independent variables in the clinical group. Cronbach’ alpha to determine the test-retest reliability.

In sum, the results of the present study indicated that feeding problems were significantly higher in the oral, pharyngeal and esophageal phases for the clinical group in comparison to the control group. The feeding problems in children with Down syndrome were highest in the oral phase followed by pharyngeal and esophageal phase. Within the oral and the pharyngeal phases of swallow the feeding difficulties were significantly greater for solids than liquids. It was also found that parent’s/caregivers lacked awareness of feeding difficulties faced by their children. The feeding problems in

the clinical group could be attributed to the oromotor problems which were present to a significantly greater extent than the control group. Further, the tongue movement was affected to a greater extent compared to the jaw and the lip movement in the children with Down syndrome. Age wise comparison in the clinical and control group indicated higher mean scores in the lower age group (2-5 years) in comparison to the higher age group (5.1 -7 years). However no significant difference was seen. Further the duration of intervention did not influence the feeding abilities.

It can be concluded from the present study that feeding difficulties are predominantly present in children with Down syndrome. The feeding difficulties exhibited by these children could be due to immature chewing pattern, poor bolus control and manipulation, primitive forward-backward movement of the tongue against the palate, tongue thrusting, poor lateralization of tongue, pooling of food in the lateral and anterior sulcus , swallowing of large, poorly chewed morsels, esophageal obstruction and gastroesophageal reflux. In addition children with Down syndrome also faced problems in oro-motor abilities which could have impacted on feeding, eating and drinking. These poor oro-motor abilities could also be associated with oral hypotonicity leading to restricted tongue, jaw and lip movements. To add on, sensory issues such as oral hyposensitivity could have also impacted on feeding in children with Down syndrome. In addition, the typical craniofacial characteristics present in the clinical group such as class III malocclusion, tongue thrust, large tongue size and a reduced number of teeth could have further decreased the masticatory capacity.

Implications of the study

The questionnaire developed provides cutting edge information on feeding skills which will provide valuable input to the speech-language clinician during the treatment of feeding problems in children with Down syndrome. The wealth of information generously provided by the participants gives new insights about the experience of feeding children with Down syndrome. The study also highlights the importance of including feeding assessment in the evaluation protocol of infants and children with Down syndrome. The information will also help in counseling the caregivers, deciding the success or failure of feeding therapy and thereby help in predicting the prognosis of the child. Emphasis should also be laid upon on the importance of including goals to overcome attending feeding problem during intervention which will help in reducing the persistence of feeding difficulties into adulthood. The rating scale can also be used to monitor the progress achieved during feeding therapy by comparing the pre-therapy with the post-therapy scores. The lack of parent/caregiver awareness of child's feeding difficulties should also be targeted by counseling parents, providing them with brochures on normal feeding development and by conducting public awareness programs on early identification and intervention of feeding problems. The present study also emphasizes on early assessment and intervention as feeding difficulties can persist into adulthood.

Limitations of the study

The study could be carried out on a geographically larger population of children with Down syndrome. The questionnaire could also include a domain on assessing apraxia in children with Down syndrome and its effect on feeding.

Future Directions

Longitudinal studies on feeding difficulties in children with Down syndrome could be carried out in order to study the developmental change in feeding in greater detail. Wider age range of children could be selected for the study. Feeding problems can be assessed in other syndromic conditions also. An objective study using equipments can provide more detailed information on the feeding and swallowing difficulties.

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Appendix

QUESTIONNAIRE TO ASSESS FEEDING PROBLEMS IN CHILDREN WITH DOWN SYNDROME

SECTION I: DEMOGRAPHIC DATA & GENERAL HISTORY

Name of the child:

Registration No:

Age/ Gender:

Date of Birth:

P.D:

Date of evaluation:

Present address:

Permanent address:

Mobile no:

Landline no (if any):

Email id

I. DEVELOPMENTAL HISTORY:

Gross Motor Skill	Speech and Language Skill
Neck control :	Babbling:
Sitting:	1 st word:
Walking:	1 st sentence:
Overall impression -Delayed /Normal	Overall impression -Delayed /Normal
Fine Motor Skill	
Palmar grasp:	
Pincer grip:	
Tripod grip:	
Overall impression -Delayed /Normal	
Present weight and height:	Birth weight:

II. DETAILS OF DIFFERENT EVALUATIONS

1. Speech and language evaluation:		
Test used:	Test results:	Interpretation:
Oromotor Reflex:		
Gag (Normal/Pathological)		

Bite	(Normal/Pathological)
Rooting	(Normal/Pathological)
Sucking	(Normal/Pathological)
Transverse tongue reflex	(Normal/Pathological)
Tongue protrusion	(Normal/Pathological)
2. Psychology evaluation:	
Test used:	Test results: Mental/Developmental Age: Social Age: Intelligence Quotient:
3. Physiotherapy/ Occupational therapy evaluation:	
Tone :	(Normal/ Hypotone / Hypertone)
Strength:	(Normal/ Affected)
Activities of Daily Living:	(Normal/ Affected)
Gross Reflex:	(Normal/ Pathology)

Associated Problems:	Yes	No
* Visual impairment. If yes specify_____		
* Any corrective measures taken		
* Hearing impairment. If yes specify_____		
* Any corrective measures taken		
* Seizures. If yes specify_____		
* Attention/ behavioral problems.		
* Hypo/hypersensitivie to touch/smell/auditory stimulus.		
* Respiratory issues such as		
– Aspiration		
– Bronchitis or chronic upper respiratory infection		
– Allergies or asthma		
– Noisy breathing: (With feeds/ other times)		
– Trouble breathing during feeds		
– Mouth breathing		
– Others		
* Heart / kidney problems.		
* Any other medical problems, If yes specify_____		

Therapy details: Type of therapy/Treatment taken	Yes/No	Duration & Frequency/Week
* Speech therapy		
* Physical therapy		
* Occupational therapy		
* Behavioural therapy		
* Surgical		
* Pharmacological		
* Feeding therapy		
* Dietician / Nutritionist		
* Special Education		

SECTION II: OROSENSORIMOTOR ASSESSMENT

1. CRANIOFACIAL ABNORMALITIES:

Sl.No		Yes	No
1.	Is the jaw size abnormal? If yes specify(too small/too large)		
2.	Is the lip size abnormal? If yes specify(too small/too large)		
3.	Is there a cleft in the lip? If yes specify(unilateral/bilateral)		
4.	Is the tongue size abnormal? If yes specify(large/small)		
5.	Is tongue thrust present?		
6.	Is the tongue fissured?		
7.	Is the arching of the palate affected? If yes specify(low, high)		
8.	Is the soft palate short?		
9.	Is there a cleft in the palate? If yes specify(unilateral / bilateral)		
10.	Is the uvula deviated? If yes specify(right/ left)		
11.	Is the uvula bifid?		
12.	Is the dentition (number/placement of the teeth) affected?		
13.	Is the dentition affected such as:		
14.	* Openbite (Upper incisors do not cover lower incisors)when jaws are closed		
	* Overbite (Upper incisors too far anterior relative to lower incisors)		
	* Underbite (Lower incisors overlaps upper teeth)		
	* Crossbite (abnormal jaw position)		
	* Supernumerary teeth		
	* Missing teeth		

	* Wide spaces between teeth		
	* Tooth decay		
15.	Is teeth grinding present? If yes(sometimes/ all the time)		

2. ORAL HYGIENE:

Sl.No		Yes	No
1.	Does the child have poor oral hygiene?		
2.	Does the child frequently get ulcers?		
3.	Is there plaque and debris accumulation?		

3. SENSORY ASSESSMENT:

Sl.No		Yes	No
	SENSORY ASSESSMENT		
1.	Is it difficult for the child to perceive the sensation of light touch on lips? If yes specify(upper/lower)		
2.	Is it difficult for the child to perceive the sensation of light touch on tongue? If yes specify(anterior/ middle/posterior)		
3.	Is it difficult for the child to perceive the sensation of light touch on cheeks? If yes specify(right/left)		
4.	Is it difficult for the child to perceive the sensation of light touch on the gums? If yes specify(upper/lower)		
5.	Is it difficult for the child to perceive the sensation of light touch on the palate? If yes specify(hard/soft)		
6.	Is it difficult for the child to perceive the temperature variations on the lips?		
7.	Is it difficult for the child to perceive the temperature variations on the tongue?		
8.	Is it difficult for the child to discriminate the differences in taste? If yes specify(sweet/sour/salty/bitter)		
Total			

SECTION III: FEEDING ASSESSMENT

I. GENERAL FEEDING ISSUES			
Sl.no		Yes	No
GF.1	Does your child have feeding problems? If yes, at what age did you notice the problem?		
GF.2	Describe your child's feeding problem:		
GF.3	Are you concerned about your child's feeding problem?		
		Very	

	If yes, specify	much concerned	
		Fairly concerned	
		Not concerned	
GF.4	Are you concerned about the child's weight? If yes, specify	Very much concerned	
		Fairly concerned	
		Not concerned	
GF.5	Is feeding problem present for all types of food?		
	* Semisolid		
	* Solid		
	* Liquid		
	Is this problem consistently present?		
GF.6	Do main meals take a long time? If yes, how long?		
	* <1/2 hr		
	* 1/2 hr		
	* 1/2 hr-1 hr		
	* 1-2 hr		
	* >2hrs		
GF.7	Do you need to force the child to eat? If yes specify		
	* Verbally		
	* Physically		
GF.8	Does the child have irregular bowel movement? If yes specify		
	* Every other day		
	* Once in 2-3 days		
	* Others _____		
GF.9	Does the child have any food allergy? If yes specify		
GF.10	Is the child receiving any special diet (Kosher, gluten-free, etc.)? If yes specify		
GF.11	Is the child dependent on you for feeding? If yes specify,		
	* Completely dependent		

	* Partially dependent		
	* Independent		
GF.12	Does the child have a poor appetite?		
GF.13	Does the child show these symptoms during a mealtime?		
	* Refuse to eat		
	* Does not respond to food.		
	* Messy eater		
	* Over eats		
	* Eats little		
	* Eats a limited variety of food/ selective		
	* Difficulty progressing to table food.		
	* Others		
GF.14	Does the child show these behavior problems during mealtime?		
	* Tries to get out of seat		
	* Cries/screams		
	* Falls asleep		
	* Spits out food		
	* Throws food/utensils		
	* Leaves the table before finished		
	* Takes food from others		
	* Aversive behaviour to oral tactile input		
	* Others		
GF.15	What is the child's position during mealtimes?		
	* Infant seat		
	* Child stands		
	* Child wanders around		
	* In front of TV		
	* Held in caretaker's arms		
	* On caretaker's lap		
	* On floor.		
	* Regular chair and table		
	* Slightly Reclined position		
	* Completely reclined position		
	* Adaptive chair Type: _____		

II. FEEDING HISTORY

Sl.no		Yes	No
FH.1	Did the child have difficulty with breast feeding?		
	Did the child have difficulty with sucking from a bottle?		
	At what age was bottle feeding introduced?		

FH.2	Was difficulty seen in transition from breast feeding to bottle feeding?		
FH.3	Did the child have difficulty eating baby food from spoon? If yes, please explain _____ At what age was spoon feeding introduced _____		
FH.4	Did the child have difficulty in drinking from a cup/glass? If yes, please explain		
	* Bites cup		
	* Single sip/consecutive sip		
	* Swallowing difficulty		
	* Loss of liquid		
	* Others:		
	At what age cup/glass drinking was introduced? _____		
FH.5	Did the child have difficulty in eating independently (pieces of chapatti/dosa /idli)? At what age was this noticed?		
	Did the child have any of these problems?		
	* Biting difficulty		
	* Chewing difficulty		
	* Swallowing difficulty		
	* Others		
FH.6	Did the child have difficulty in eating independently with spoon? At what age was this seen?		
FH.7	Did the child have difficulty in drinking through a straw? If yes specify _____		
	* Bites straw		
	* Drinks only at a particular angle – Near Horizontal – Vertical – 45 degree		
	* Loss of liquid		
	* Difficulty in drinking – Thick liquids – Thin liquids		
	* Swallowing difficulty		
	* Others:		
	At what age was straw drinking seen? _____		

III. MODIFICATIONS DURING FEEDING

Sl.no		Yes	No
MF.1	Is there any modification made to the utensils used for feeding? If yes, specify the modification done for		
		Nipple:	
		Plate:	
		Spoon:	
MF.2	Is there any modification done in the seating, If yes specify, * Floor/ Bed * Lap; Supine position(Chair / Floor/ Bed)		
		Head supported	
		Head not supported	
	* Regular chair and table		
	* Slightly Reclined position		
	* Completely reclined position		
	* Adaptive chair Type: _____		
MF.3	Food consistency: what is the consistency of food that is currently applicable?		
	Food consistency		
	* Liquids/soups		
	* Blenderized table food		
	* Mashed table food		
	* Regular table food		
	* Crisp foods (chips, muruku)		
	* Chewy foods (meat, halwa)		
	* Crunchy foods (carrots)		
MF.4	Is any alteration made to temperature to suit child's needs? If yes specify, Preferred food temperature		
	* Warm.		
	* Luke warm		
	* Cold		
	Preferred liquid temperature:		
	* Warm		
	* Luke warm		
* Cold			

MF.5	Is any support given to jaw/lip control? If yes specify,		
	* Support some of the time		
	* Support all of the time		
MF.6	Is support provided for self-feeding? If yes specify,		
	* Child encouraged to be involved in self-feeding-few mouths		
	* Child encouraged to be involved in self-feeding for a mouth.		
	* Support provided on :	Palm:	
		Wrist:	
	Elbow:		
	Shoulder:		
MF.7	Is the quantity of food restricted (only small amounts given) per mouthful?		
MF.8	Any alternative feeding method used? If yes specify,		
	* Nasogastric/gastric tube feeding		

Any other:

SECTION IV: ASSESSMENT OF DIFFERENT PHASES OF SWALLOW

1. ORAL PREPARATORY AND ORAL PHASE (✓ the options)

Scoring: Yes: 1, No: 0

Items	SOLID		LIQUIDS		Remarks if any
	Yes	No	Yes	No	
1. Is it difficult to open jaw to receive the food?					
2. Is jaw closure absent after receiving the food?					
3. Is the head alignment abnormal while receiving food?					
4. Is difficult to form lip seal during bolus formation/ liquid intake?					
5. Is it difficult to form a bolus?					
6. Is it difficult to control the bolus?					
7. Is difficult to move the tongue laterally during bolus preparation?					
8. Is it difficult for the child to bite food items?					
9. Is it difficult to move the upper lip during spoon feeding?					

10. Is there lack of awareness of food in the mouth?					
11. Is the flow rate poor (bubbles with each suck)?					
12. Does the child retain food in the mouth without chewing?					
13. Is it difficult for child to chew food?					
14. Does it take long duration for bolus manipulation/ chewing?					
15. Is it difficult for the child to retain food/liquid within the oral cavity?					
16. Is anterior-posterior tongue movement/tongue peristalsis difficult?					
17. Is the tongue coordination difficult?					
18. Is the oral transit time inappropriate?					
19. Is coughing present before swallowing?					
20. Does the child have trouble breathing normally during bolus manipulation?					
21. Is the suck/swallow- respiratory sequence incoordinated?					
Total					

Examiner's rating:

Please rate severity of the problem faced during oral preparatory and oral phase on the scale below:

Severity	None	Mild	Moderate	Moderately Severe	Severe
Rating	0	1	2	3	4

2. PHARYNGEAL PHASE: (✓ the options)

Scoring: Yes: 1, No: 0

Items	SOLID		LIQUIDS		Remarks if any
	Yes	No	Yes	No	
1. Does the child retain food in the mouth after chewing without swallowing?					
2. Is there a delay in swallowing?					
3. Does the tongue thrust out during swallowing?					
4. Is lip seal absent during swallowing?					
5. Is aspiration /choking during liquid intake seen?					
6. Does food pool in the anterior sulcus?					
7. Does food pool in the lateral sulcus?					
8. Is nasal regurgitation seen?					

9. Is gag reflex produced during swallow?					
10. Does the child take multiple swallows per bolus?					
11. Does the child complain of discomfort in the throat during swallowing?					
12. Does the child raise head/hyper extends while swallowing?					
13. Is there no elevation of larynx during swallowing?					
14. Does the child clear throat during/after swallow?					
15. Does the child cough food material after swallow?					
16. Does the child take sips of water after every swallow?					
17. Is the child not aware of the residue in the mouth?					
18. Is gurgly voice seen : -During feed? -After feed?					
19. Is swallowing effortful?					
20. Is any sign of distress seen after swallow?					
21. Are there changes in face during swallow -Color -Tear in eyes					
Total					

Examiner's rating:

Please rate severity of the problem faced during pharyngeal phase on the scale below:

Severity	None	Mild	Moderate	Moderately Severe	Severe
Rating	0	1	2	3	4

3. ESOPHAGEAL PHASE (✓ the options)

Scoring: Yes: 1, No: 0

Symptoms	Yes	No	Details/remarks (if any)
1. Does the child indicate of food getting stuck in lower throat or chest?			
2. Does the child frequently vomit after feeding?			
3. Does the child complain of burning sensation in mouth or throat after feeding(esophagitis)?			
4. Does the child get gagging sensation towards			

the end/after meals?			
5. Does the child awaken at night with gagging/coughing?			
6. Does the child get regurgitation after lying down?			
Total			

Examiner's rating:

Please rate severity of the problem faced during esophageal phase on the scale below:

Severity	None	Mild	Moderate	Moderately Severe	Severe
Rating	0	1	2	3	4

MAIN FEEDER'S OVERALL SEVERITY RATING OF FEEDING

Severity	None	Mild	Moderate	Moderately Severe	Severe
Rating	0	1	2	3	4

EXAMINERS'S OVERALL SEVERITY RATING OF FEEDING

Severity	None	Mild	Moderate	Moderately Severe	Severe
Rating	0	1	2	3	4