Videofluoroscopic evaluation of swallowing in bottle fed infants and children with laryngomalacia

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Abstract

Purpose: Laryngomalacia is a congenital anomaly characterised by inward prolapse of supraglottic structures during inspiration. This study explores the impacts of laryngomalacia on swallowing biomechanics using quantitative measures from videofluoroscopic swallow studies (VFSS).

Method: A retrospective clinical audit at one tertiary hospital (2012–2022) identified 877 children ascribed a diagnostic code for laryngomalacia, with 228 (26%) seen by speech language therapy and 26 (3%) receiving a videofluoroscopic swallow study (VFSS). Six VFSS were excluded due to method of fluid intake, non-compliance, study quality or coding error. The VFSS of 20 children (aged 1–23 months; 12 male) diagnosed with laryngomalacia were analysed: six with laryngomalacia in isolation; six with a co-existing chromosomal or neurological disorder, e.g., Beckwith-Wiedmann syndrome, and eight with co-existing additional anatomic abnormality, e.g., pectus excavatum or base of tongue collapse. Five children had tracheostomy insertion and 12 underwent supraglottoplasty (nine prior to their VFSS study). Studies were analysed for the presence of penetration or aspiration, pharyngeal residue, and retrograde bolus movement. Quantitative timing and displacement measures were collected and compared to previously published values indicating risk of airway violation or bolus retention in bottle fed infants.

Results: Thirteen out of twenty children aspirated during VFSS (ten of whom aspirated silently). Children with laryngomalacia in isolation had significantly longer times to achieve airway closure (Airwaycl) in comparison to children with concurrent medical conditions ($H = 6.810$, $p > .05$). Longer times to achieve airway closure (Airwaycl) were correlated with increased Penetration-Aspiration Scale (PAS) scores (Max PAS $R_s = 0.588$, p .01). Delayed timing of airway closure in relation to bolus reaching the pharyngoesophageal segment (PES)

(BP1AEcl) also correlated with increased PAS scores (Max PAS $R_s = 0.648$, $p < .01$). Total pharyngeal transit times (TPT) were longer in all groups when compared with previously published 'at risk of aspiration' thresholds. Pharyngeal constriction ratio (PCR) was elevated (≥ $0.1cm^2$) in two children (0 = laryngomalacia in isolation; 1 = syndromic; 1 = anatomic). Sixty-five percent of children were referred for VFSS after surgery, with ten out of thirteen of the aspirators in the post-supraglottoplasty group. Seven children out of 20 received a follow-up VFSS, of which three VFSS assessed bottle feeding, limiting conclusions that could be drawn about change over time.

Conclusions: Only a small number of children diagnosed with laryngomalacia are referred for VFSS. Of these, the majority are post-supraglottoplasty and many have multiple comorbidities alongside laryngomalacia. Aspiration, prolonged transit times and 'at risk' airway closure timings are common in those with laryngomalacia in isolation as well as those with other comorbidities, even after supraglottoplasty. While VFSS may not be necessary for all children with laryngomalacia, instrumental assessment should be considered, especially in those with feeding difficulty, respiratory concerns or other comorbidities.

Preface

This master's thesis conforms to the referencing style advised by the American Psychological Association Publication Manual (7th ed.) and spelling advised by the Oxford English Dictionary.

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My clinical experience in the field of paediatric dysphagia began in 2020, and special thanks go to my colleagues and educators who allowed me to develop skills in assessment and therapy and develop my passion for infant feeding. I would also like to thank Waitematā Child Development Service for their flexibility and support.

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List of Abbreviations

ASHA	American Speech-Language-Hearing Association
FEES	Fibreoptic endoscopic evaluation of swallowing
GERD	Gastroesophageal reflux disease
HFNC	High flow nasal cannula
IPOG	International Paediatric ORL Group
LFNC	Low flow nasal cannula
MDT	Multidisciplinary team
MLB	Microlaryngobronchoscopy
NAS	Neonatal abstinence syndrome
nCPAP	Nasal continuous positive airway pressure
NGT	Nasogastric tube
NICU	Neonatal Intensive Care Unit
ORL	Otorhinolaryngology
PAS	Penetration–Aspiration Scale
PEG	Percutaneous endoscopic gastrostomy
PFD	Paediatric feeding disorder
PES	Pharyngoesophageal segment
SALs	Synchronous airway lesions
SCBU	Special Care Baby Unit
UES	Upper oesophageal segment
VFSS	Videofluoroscopic swallow study

Chapter 1 Introduction

Feeding is a complex process that involves the interaction of the central and peripheral nervous system, oropharyngeal structures, cardiopulmonary system, and gastrointestinal tract with support from a skeletal system (Goday et al., 2019). Disruption of one of these systems can lead to the development of a paediatric feeding disorder (PFD) which occur in many paediatric diseases and disabilities. PFDs result in impaired oral intake, requiring a multidisciplinary approach to intervention. One specific paediatric population with a well-documented prevalence of PFD is children with laryngomalacia (Chadha, 2019). Laryngomalacia is a congenital anomaly of the larynx typically arising in infancy and is self-limiting in nature within the first five years of life (Bedwell & Zalzal, 2016; Hilland et al., 2016). The international literature reports a clear association between laryngomalacia and swallowing dysfunction, with 49% of cases experiencing aspiration events thought to be related to either the disruption of the suck, swallow, breath pattern required for infant feeding (Jaffal et al., 2020) or atypical laryngeal sensorimotor integrative function (Thompson, 2007).

This study aims to describe the swallow physiology of paediatric bottle fed patients with laryngomalacia using retrospective videofluoroscopic swallow study (VFSS) data. A literature review was conducted that focused on neonates (≤28 days old) and infants (> 28 days to ≤12 months) with laryngomalacia, as this study solely assesses bottle fed children. For simplicity, for the remainder of this thesis, the term 'child' describes the full spectrum of childhood from neonates through to adulthood and 'infant' describes children under 12 months of age. This literature review provides an overview of the consensus components of the paediatric clinical feeding assessment, followed by a critical appraisal of the body of evidence currently available on feeding and laryngomalacia. The study rationale and design are described, and findings are presented and discussed in the context of the current literature and clinical practice when caring for children with PFDs.

Paediatric Feeding Disorders

PFDs are defined as "impaired oral intake that is not age-appropriate, and is associated with medical, nutritional, feeding skill, and/or psychosocial dysfunction" (Goday et al., 2019, p. 124). The diversity of conditions associated with feeding difficulties led researchers to create this overarching and unifying diagnostic term. The term was proposed to promote and advance research by creating a critical knowledge base and directing healthcare policy and clinical evidence-based practice (Goday et al., 2019). PFD acknowledges that feeding is a complex process in which the four domains can be affected simultaneously or as a secondary complication of each other (Goday et al., 2019). Therefore, it is unsurprising that PFD presentations are heterogeneous, and occur in relation to multiple different diseases and disorders, including paediatric cardiac conditions, neurogenic disorders such as cerebral palsy or vocal fold palsy, or structural difficulties such as those found in Beckwith–Weidemann syndrome or DiGeorge syndrome (see Table 1).

Table 1

Paediatric aetiologies associated with feeding problems (Miles, 2019)

- **Age-related complications:** Prematurity
- **Central nervous system disorders:** Cerebral palsy, neurologic abnormalities, tumours
- **Cranial nerve, peripheral nerve and muscle disorders**: Moebius syndrome, muscular dystrophies, spinal muscular atrophy
- **Chromosomal abnormalities:** Trisomy 21, Beckwith–Weidemann syndrome
- **Genetic structural conditions**: Pierre Robin disorder, cleft palate
- **Metabolic disorders:** Hypoglycaemia, pituitary and hypothalamic disorders, foetal alcohol syndrome
- **Gastrointestinal diseases:** Strictures, hiatal hernia, gastroesophageal reflux
- **Cardiorespiratory compromise:** Congenital heart disorders, chronic lung diseases
- **Inflammatory diseases:** Children in pain with arthritic disorders, juvenile dermatomyositis
- **Medication side effects:** Hypotonia, drowsiness, nausea from oncology medications
- **Sensory deprivation/overstimulation:** Prolonged acute hospitalisations, autism spectrum condition

PFD can be categorised by an underlying diagnosis, specific dysphagic impairment or onset presentation: either an *acute* presentation (ICD-11 code R63.31; World Health Organization [WHO], 2021) in which feeding difficulties have been present for less than 3 months or *chronic* feeding difficulties (ICD-10 code R63.32; WHO, 2021) where difficulties were present for 3 or more months. PFD presents differently depending on the underlying cause and the child's age. PFD difficulties can also categorised into feeding difficulties where deficits impact all aspects of eating and drinking at all stages of the swallow as well as the social experience (Arvedson & Brodksy, 2002); alternatively, swallowing difficulties refer to the biomechanical transit of liquids or food from the oral cavity to the stomach(Logemann, 1998). PFD may present at several stages of the swallow: A) *oral phase* difficulties which may be related to structural anomalies such as cleft lip or palate, micrognathia, or macroglossia; B) *pharyngeal stage* difficulties associated with respiratory disease, prematurity, cardiac conditions, or pharyngeal structural anomalies; and C) *oesophageal stage* difficulties commonly associated with atresia, fistulas, or strictures. Nerve damage and neurodevelopmental disorders can affect all three stages of the swallow. PFD can also be a skills-based dysfunction encompassing delayed or inefficient oral skills, with not all PFD cases demonstrating airway violation events. Refusal behaviours associated with generalised feeding difficulties can be due to a history of aversive feeding practices (e.g., enteral feeding or a choking event) or sensory difficulties, as seen in neurodevelopmental conditions such as autism spectrum condition.

Prevalence of Paediatric Feeding Disorders

The prevalence of PFD is thought to be as high as 1:23 in children who are under 5 years old (Kovacic et al., 2021). Other estimates suggest that PFD rates are as high as 85% in children with disabilities and up to 5% in otherwise healthy children (Dodrill, 2020). With advancements in medical care, there has been an increase in survival rates for children with congenital abnormalities and infants born at younger gestational ages. Studies have stated that there may be

a correlation between the steady increase in children diagnosed with PFD and increased infant survival rates, although definitive statistics are not available (Estrem et al., 2016; Silverman et al., 2021). PFD in preterm infants often emerges during the first year as feeding becomes volitional and the oro-motor demands increase (Connell et al., 2023; Pados et al., 2021).

Swallowing and Airway Violation

There are a range of biomechanical differences in the neonate to allow for uncompromised feeding. Here, I will briefly summarise the process of swallowing in an unaffected infant during breast or bottle feeding, to allow for subsequent comparison and evaluation of feeding in PFD cases. Feeding is a sensorimotor process and neurobehavioral skill that relies on maturation (Jadcherla, 2017; Ross & Fuhrman, 2015). The anatomic structures involved in the swallow are the lips, tongue, jaw, palate, hyoid bone, pharynx, larynx and oesophagus (Wolf & Glass, 1992), and there are 26 muscles controlled by the cortex and brainstem that transport material to the stomach (Jadcherla, 2017). The anatomical changes to the oral cavity, pharynx, and larynx over the first three years support safe oral feeding and texture progression (Morris & Klein, 2000).

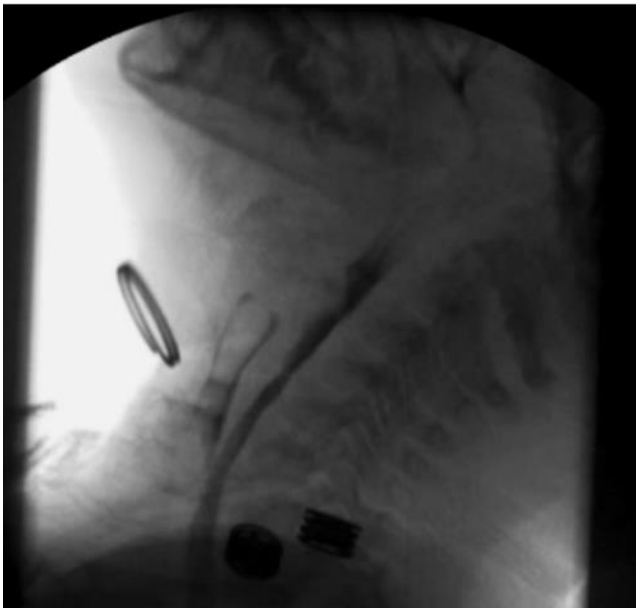
Two sucking patterns present in infancy are non-nutritive and nutritive sucking, both of which should be rhythmical, timely, and coordinated (Foster et al., 2016; Morris & Klein, 2000). Non-nutritive sucking is involved in the management of saliva, is a self-soothing activity, and typically occurs at twice the rate of nutritive sucking (Lau et al., 2003; Wolff, 1968). Nutritive sucking is a sensorimotor activity that comprises the coordination of sucking, swallowing, and breathing for the purposes of liquid ingestion (Ross & Fuhrman, 2015). During nutritive sucking, the bolus is transported from the teat into the oral cavity using compression and suction creating an intra-oral vacuum (Lagarde et al., 2021; Ross & Philbin, 2011). Sucking pads and jaw differences in infancy provide stability for early feeding. The repetitive tongue motion is a primitive reflexive pattern in early infancy which typically diminishes between 4 and 6 months

of age as the infant moves from reflexive to volitional feeding. The bolus is held in the oral cavity and the tongue rises whilst the soft palate, sealed posteriorly, prevents the bolus escaping through the nasopharynx. The bolus is subsequently propelled into the pharynx when the base of tongue retracts. In infants, the larynx is positioned higher in the pharynx and under the base of the tongue, providing increased airway protection compared to an older child or adult (Delaney & Arvedson, 2008). The larynx elevates and the pharyngeal walls shorten. The epiglottis closes over the arytenoids, the vocal folds close, and there is a pause in respiration to protect the airway. The bolus is propelled through the pharynx towards and through the pharyngoesophageal segment (PES) to the upper oesophageal sphincter (UES) (Wolf & Glass, 1992). Opening of the UES and constriction of the pharyngeal constrictors helps move the bolus through into the oesophagus.

Penetration or aspiration may occur when the timing, coordination or mechanics of this process is altered. Rosenbek et al. (1996) clearly defined both penetration and aspiration and proposed the Penetration Aspiration scale (PAS) which provided a graduated rating scale reference for professionals to describe and quantify airway violation. *Penetration* is defined as the passage of material (bolus) into the larynx that does not pass below the vocal folds, whereas *aspiration* (see Figure 1) is defined as bolus passing below the level of the vocal folds (Rosenbek et al., 1996). PAS 3 or greater scores are typically considered airway violation (Daggett et al., 2006; Steele & Grace-Martin, 2017) and present a risk to long-term respiratory health.

Figure 1

A videofluoroscopic image in lateral view of a 16-month-old child swallowing barium, demonstrating barium in the airway anteriorly (PAS 8 = silent aspiration) and in the pharynx and oesophagus posteriorly



Signs and Symptoms of Paediatric Feeding Disorders

Dysphagia in infants may be identified through adverse cardiorespiratory events, including apnoea, bradycardia, oxygen desaturation or increased work of breathing, and cyanosis or stress cues. In older children, swallow difficulties may present as adverse mealtime events, such as coughing, delayed oro-motor skills, gagging, vomiting, fatigue, or refusal (Weir et al., 2007). When identifying penetration or aspiration events in children, wet vocal quality, wet breathing, or cough are the most predictive clinical signs of aspiration at bedside (DeMatteo et al., 2005; Silva-Munhoz et al., 2015). Accurate diagnosis of swallowing impairment however, requires instrumental examination, as by definition, silent aspiration, cannot be identified at bedside (Arvedson et al., 1994).

Long term complications of PFD may include malnutrition, dehydration, faltering growth, respiratory complications, as well as familial and child distress (Lefton-Greif & McGrath-Morrow, 2007; Loughlin, 1989). The specific long term consequences of airway

violation (recurrent chest infections, tracheal and bronchial granuloma, stenosis, bronchitis, bronchiectasis, empyema, or respiratory failure) are life-limiting and require investigation and management in the paediatric context given the need for the respiratory system's longevity (Tutor & Gosa, 2012).

Feeding difficulties are associated with high levels of parental stress (Didehbani et al., 2011), which can be thought of as a secondary consequence or symptom of PFD. Children with PFD often have comorbidities that impact development, creating a strong parental priority to deliver nutrition to support neurodevelopmental potential (Estrem et al., 2016). However, this feeding-related parental stress is not unique to parents of children with atypical development. UNICEF recognises the role of parental stress in relation to suboptimal feeding styles in typically developing children under 5 years old, with restricted feeding practices, either pressure to offer or restrict food, cited as the main consequence (Almaatani et al., 2023). Specifically, for parents of children with PFD, negative child behaviours and lower child developmental function increase parental stress, distress, and parent-child dysfunctional interactions (Silverman et al., 2021). While food refusal or nutritional concerns cause stress to all parents, parents of children with PFD have specific and unique concerns related to their child's development as well as nutritional status.

Management of Paediatric Feeding Disorders

PFD is a multi-system failure; therefore, a multidisciplinary team (MDT) is required for effective and safe management. In a systematic literature review of intensive multidisciplinary interventions for children with PFD, core disciplines involved included psychology, nutrition, medicine, and allied health, specifically speech pathology or occupational therapy (Sharp et al., 2017). The MDT may also include physiotherapy, lactation consultants, and clinical nurse specialists (Dodrill, 2020; McComish et al., 2016). The medical team is typically separated into primary care and specialist medical care (Dodrill, 2020); medical specialists include paediatric

otolaryngologists, gastroenterologists, developmental paediatricians, neurologists, pulmonologists, cardiologists, radiologists, and psychiatrists (Miles, 2019).

PFD is often not an isolated difficulty, and it is vital that the MDT communicates effectively to create clear and timely shared goals. The *International Classification of Functioning, Disability and Health* framework (WHO, 2021) is useful when assessing a child to create holistic, family, and person-centred care goals. The framework aids professionals in deciding the healthcare priority; for example, there may be times in the acute setting (NICU/ SCBU) when the MDT deems supplementary oxygen weaning to be a higher priority than nasogastric tube (NGT) feed weaning. Regular MDT communication is essential for competent and coherent assessment and management. Treatment for PFD mainly focuses on safe oral intake, increased oral intake, improved mealtime behaviours, and reduced parental stress (Sharp et al., 2017). When considering supporting infant feeding, infant-driven feeding practice is routinely implemented in treatment, which assesses an infant's readiness for feeding and caregiver strategies (Ludwig & Waitzman, 2007; Ross & Philbin, 2011).

Clinical Feeding Examination

Assessment often begins with screening through a referral process. Appropriate referrals for a clinical feeding assessment include nutritive sucking difficulties in an infant, overt signs of aspiration (i.e., changes in respiration, eye watering, nasal flaring, gagging, choking, or coughing), unexplained food/fluid refusal, faltering growth, recurrent pneumonia/chest infections, lengthy mealtimes, drooling, reflux/vomiting or high-risk dysphasic medical diagnoses such as trisomy 21, cerebral palsy, or cleft palate (Arvedson, 2008; Samour & King, 2006).

If a clinical feeding assessment is warranted, a caregiver case history will be completed to gather pertinent information regarding medical, growth, diet, nutritional status, early feeding, general development, and behaviour concerning mealtimes. This history is followed by a

feeding assessment guided by the child's age and developmental stage. A clinician would be expected to complete a physical examination, assessing development (including age-related reflexes associated with feeding), respiratory status, and oral anatomy (American Speech-Language-Hearing Association [ASHA], n.d.). Consideration should also be given to appropriate and expected developmental motor skills and muscle tone (i.e., hypertonia, hypotonia, or mixed tone), as motor skills and muscle tone influence the ability to feed independently and safely.

If the patient is an infant, determination of oral feeding readiness should be completed (see Table 2). If the infant is not in an optimal state for feeding, the feeding assessment should be delayed, with priority given to developing the infant's medical stability or improving state and behavioural organisation.

Table 2

Stability required for oral feeding

Readiness for oral feeding	
Physiologic maturation	Digestion, respiration, heart rate, and oxygenation range
Motor	Muscle tone, motor control, midline movements
Behavioural organisation states	Ranging from sleepy, drowsy, *quiet/actively awake, highly aroused/agitated

Note. *Optimal state for oral feeding. Adapted from ASHA (n.d.) and Delaney and Arvedson (2008).

Assessment of oro-motor skills in relation to non-feeding tasks (i.e., non-nutritive sucking/oro-motor exam) and feeding tasks (i.e., nutritive sucking/mastication), observation of pharyngeal stage of swallow, and environmental factors that affect mealtimes are also expected (Dodrill, 2020). Environmental factors encompass caregiver interactions in relation to feeding and communication. Following this, therapeutic strategies and goal setting should be considered, which are discussed later in this review.

There are both standardised and non-standardised assessments published for children who have symptoms of feeding or swallowing difficulties. These include screening tools, observational checklists, assessment protocols, and parent questionnaires (Miles, 2019). A recent systematic review of screening tools identified 44 published screening assessments that effectively detect generic PFDs (Litchford et al., 2021). Many studies also offer critical reviews of assessments designed for specific populations within PFD, such as those with cerebral palsy (Benfer et al., 2012). Standardised infant feeding assessments and consensus protocols that target readiness for oral feeding include The Neonatal Oral-Motor Assessment Scale (NOMAS) (Braun & Palmer, 1985), Revised NOMAS (da Costa et al., 2016), Early Feeding Skills Assessment (EFS) (Thoyre et al., 2005, 2018), Feeding Readiness Scale (Ludwig & Waitzman, 2007), Supporting Oral Feeding in Fragile Infants (SOFFI) (Ross & Fuhrman, 2015), Infant-Driven Feeding Scales (IDFS) (Waitzman et al., 2014) and the Neonatal Eating Assessment Tool (NeoEAT) (Pados et al., 2016, 2019). These assessments provide clinicians with a framework to complete assessment or gather further information from parents, but further instrumental assessment is sometimes required to ascertain the presence or severity of airway violation events.

Instrumental Assessment

Onward referral for instrumental assessment by VFSS or flexible endoscopic evaluation of swallowing (FEES) would be indicated when objective evaluation of swallow function is required to detect aspiration and guide management or when there is concern for potential airway violation. Symptom onset and severity, expected disease progression, and unexplained or new respiratory difficulties are all factors that should guide clinicians when identifying candidates for instrumental assessment (Dodrill, 2020).

VFSS is widely agreed to be the gold standard of instrumental assessment when assessing all phases of the swallow (Arvedson & Lefton-Greif, 2017; Hiorns & Ryan, 2006).

Objective and quantitative measures have been published to assess paediatrics VFSS at high frame rates without increasing radiation dose (Henderson et al., 2016). However, there is a need for standardisation of VFSS due to excessive variability seen in practice (Slovarp et al., 2018) especially due to the carcinogenic risks from radiation exposure (Bonilha et al., 2018, 2019). The implications of radiation dose received during VFSS are dependent on age, gender, and organs exposed. The orbit of the eyes and thyroid glands are more sensitive to radiation exposure, and infants exposed to radiation are at increased risk of thyroid cancer (Bonilha et al., 2018). Screening protocols for VFSS that exclude the eye where possible are preferable but depend upon age. It is imperative that MDT use VFSS with a clear goal for the procedure and gain the most information possible.

Due to the carcinogenic risk, the need for non-radiological assessment has been acknowledged since the 1980s (Bosma et al., 1983). Several studies have assessed the pharyngeal stage of swallowing using FEES (Mills et al., 2021; Richter et al., 2009). For example, Mills et al. (2021) assessed breastfed infants with laryngomalacia and found FEES to be an appropriate alternative for assessing breastfeeding whilst also observing pharyngeal and laryngeal anatomy. In several studies, FEES has shown greater sensitivity for the detection of penetration and aspiration when compared to VFSS, although the clinical association with FEES-graded risk is not established (Giraldo-Cadavid et al., 2017; Kelly et al., 2007). However, da Silva et al. (2010) argue that FEES should be used as a complementary assessment. FEES lacks published objective measures compared to VFSS and visualisation can be limited due to compliance as well as camera positioning. With most paediatric pharyngeal phase research in bottle fed infants reports VFSS findings rather than FEES.

When focusing on VFSS as an instrumental assessment, there is the question of overall diagnostic validity. Fluoroscopic visualisation is typically confined to the beginning of a feed due to the radiation risk. McGrattan et al. (2020) focused on pathophysiological changes in

bottle fed infants seen between 0 and 2.5 minutes and found significant differences in swallowing physiology based on timing. This finding is important with regard to clinical decision making and suggests that clinicians should consider VFSS data within the wider context of a child's respiratory health and bedside feeding observation.

Instrumental assessment allows clinicians and researchers to develop an understanding of the swallow pathophysiology of children with a range of conditions. When clinicians have clear physiologic information beyond penetration and aspiration, the variability in recommendations decreases (Kerrison et al., 2023; Slovarp et al., 2018). Ultimately, focusing on airway violation alone does not allow researchers or clinicians to understand the mechanism of aspiration, limiting the effectiveness and accuracy of recommendations for compensatory or therapeutic techniques.

Assessment Considerations – Respiratory Support

Clinicians should consider respiratory compromise and the infant's respiratory support in relation to feeding. Infants may require respiratory assistance due to respiratory complications (e.g., bronchopulmonary dysplasia) or anatomical obstruction to airflow (e.g., laryngomalacia, macroglossia etc.). Mechanical ventilation through nasal, oral, or tracheal routes and history of ventilation are critical when assessing the swallow for several reasons (Dodrill, 2020). Endotracheal intubation is associated with impaired oral motor development, altered oral sensitivity, altered palatal development, and delayed feeding development in premature infants (Enomoto et al., 2017; Poore et al., 2008). However, in a retrospective study by Alm et al. (2023) there was no significant difference between nasal and oral intubation and subsequent feeding problems. This shows that mechanical ventilation, specifically positive pressure, is a contentious issue in infant feeding both in the literature and clinical practice. This centres on the effects of positive pressure interfering with the normal movement of laryngeal structures involved in swallowing and airway protection (Gaon et al., 1999). In a recent systematic review

of 20 studies, Canning et al. (2021) conclude that there are insufficient findings to determine whether commencing oral feeding whilst on nasal continuous positive airway pressure (nCPAP) or high flow nasal cannula (HFNC) facilitates the transition to full oral feeding without adverse effects. The sole study focusing on airway protection whilst on nCPAP using VFSS was halted due to safety concerns associated with increased aspiration when compared with low flow nasal cannula (LFNC) (Ferrara et al., 2017). As thus the effect of positive pressure airflow on the laryngeal mucosal sensitivity is unknown, and any blunting of sensory function would potentially impair airway protective mechanisms.

Tracheostomies are typically placed in children with severe anatomical obstruction to airflow or when long-term mechanical ventilation is required. Swallowing deficits in children with tracheostomies are well documented due to the method affects on the sensory-motor pharyngeal stage of swallow from delayed and diminished laryngeal elevation, altered pressure systems, risk of sensory deficits and direct pressure on the oesophagus (Abraham & Wolf, 2000). Evidence for the use of speaking valves to limit aspiration events in the adult population is mixed, and in a small paediatric study, whilst speaking valves did not reduce penetration or aspiration, they did reduce residue, which may have clinical significance (Ongkasuwan et al., 2014).

Developmental Considerations

Infants and children react differently to airway violation events. Laryngeal chemoreflexes appear to be a primary sensory mechanism for defending the airway from aspiration of liquids. This typically results in apnoea or laryngeal closure in early infancy, which becomes a cough response as children develop (Thach, 2001, 2007). Whilst a premature infant is typically born with the ability to swallow, there is an absent cough reflex due to the fluid-filled lungs of a fetus in utero (Miles, 2019), and the development of full oral feeding may be delayed. A typical preterm neonate may sustain full oral feeding by 34 weeks gestation. In contrast,

infants less than 32 weeks are unable to coordinate nutritive sucking as a result of neurological immaturity and difficulty regulating autonomic functions and the state required for oral feeds (Delaney & Arvedson, 2008).

When assessing signs of aspiration in infants, clinicians should also consider the possibility of silent aspiration as a wealth of instrumental swallow data suggests silent aspiration is common in infancy (Arvedson et al., 1994; Velayutham et al., 2018). Silent aspiration is defined as material passing into the airway below the vocal folds without displaying overt response signs of coughing or discomfort (e.g., wet vocal quality, change in breathing pattern, watery eyes, cyanosis) (Rosenbek et al., 1996). In a retrospective review of children with mixed aetiology under 18 years old who underwent VFSS at a tertiary hospital, 95% of infants less than 6 months silently aspirated. Structural abnormalities were significantly associated with silent aspiration in children between 0 and 18 years old, with silent aspiration document in children with laryngeal cleft (41%), laryngomalacia (41%) and unilateral vocal fold palsy (54%) (Velayutham et al., 2018). This highlights the requirement for instrumental assessment in children with structural abnormalities.

Nevertheless, viewing aspiration within the wider context of typical feeding development and the limited normative infant swallow data that is currently available, is important. Infants are required to develop and improve feeding skills postnatally, and it is well-known that suck swallow breath coordination improves with postnatal age (Lau, 2015; Lau et al., 2003). Sucking burst duration, amplitude, and rate all decrease over time (Lang et al., 2011), and feeding duration also impacts coordination because the time between swallows increases within the first 5 minutes of feeding in typically developing term neonates (Bamford et al., 1992). Therefore, early feeding may be briefly uncoordinated with increased airway violation risk. Furthermore, in an infant animal study, all full-term piglets were found to aspirate when bolus volumes increased (Mayerl et al., 2021). Similarly, in an infant feeding questionnaire,

93% of typically developing infants coughed at least once during feeds in the first month of life (Barkmeier-Kraemer et al., 2017). Therefore, this evidence raises the question as to whether aspiration (overt or silent) in infancy is a normal and an incidental consequence of learning to feed, with the risk of aspiration increasing with bolus volume (Mayerl et al., 2021) and temporal changes (McGrattan et al., 2020).

Even if airway violation in infants is a consequence of learning to feed, it is still important to identify populations where aspiration events may be more common or the consequences more severe. Aspiration in children can interrupt normal development and cause serious long-term complications (Dodrill & Gosa, 2015). The question regarding the overall risk of aspirating fluids continues to be discussed among healthcare providers and researchers. It is acknowledged that the degree of lung trauma and infection are influenced by the acidity and volume of aspirated material, the ability to clear the airway (cough or movement), and the child's general health (Tutor & Gosa, 2012; Weir et al., 2007). Therefore, clinicians should also consider the possibility and consequences of secondary aspiration of stomach content, which is more acidic. The treatment of gastroesophageal reflux disease (GERD) will be discussed later in this review.

There is a growing movement within infant feeding to continue with oral feeds even when aspiration is a known risk because limiting oral feeding and oral stimulation impacts the development of neuromotor and sensory pathways (Desai et al., 2022). Infants aged between 0 and 6 months typically require between 420 and 840ml of feed per day to meet nutritional requirements (Samour & King, 2006), with infants who aspirate breastmilk being able to remain healthy for 3 months (Hersh et al., 2022). However, in a similar study reviewing infants longer term respiratory health over a year, infants with incidence of airway violation captured during VFSS who continued to breastfeed had significantly more respiratory based hospitalisations and had increased risk of bronchoalveolar lavage inflammation (p = .01) when compared to infants

who stopped breastfeeding (Duncan et al., 2023). These studies suggest that infants can tolerate a certain volume of aspirated fluid over a short period of time, but caution must be given to the specific types of fluids offered.

Not all children can tolerate aspiration, and in a longitudinal study, the greatest growth in the use of hospital resources was children with chronic conditions affecting two or more body systems, especially the respiratory and aerodigestive tract (Berry et al., 2013). These body systems are often implicated in swallowing disorders, and the long term adverse consequences of aspiration include recurrent chest infections, tracheal and bronchial granuloma, stenosis, bronchitis, bronchiectasis, empyema, or respiratory failure.

Continuing oral feeds in the presence of limited airway violation may support neurodevelopment. However, aspiration in medically fragile infants must be carefully managed for several reasons. First, this population typically comprises infants with limited ability to clear aspirated material or show signs of aspiration to caregivers, potentially increasing the aspiration volume. Second, due to age, respiratory health over a lifetime must be considered, as dysphagia can increase morbidity and mortality rates, especially among infants (Hernandez & Bianchini, 2019). Taking this into consideration, an MDT may coordinate a controlled, stepwise method for introducing oral feeding, including considering the child's ability to cope with aspiration, infant-driven strategies, and parental education, as an appropriate transitional method to full oral feeds (Desai et al., 2022; Ross & Philbin, 2011).

Feeding Interventions

Feeding interventions (thickened liquids, change in liquid flow rate, and/or method of liquid delivery) have been shown to improve outcomes in children with laryngeal penetration (Duncan et al., 2019). This present study focuses on bottle feeding and therefore consideration will be placed on this method of infant feeding.

Methods of Infant Feeding

Neonates and infants can typically be offered breast, bottle, or open cup for a safe fluid delivery method (Collins et al., 2004). Interestingly, the WHO's (2015) definition of exclusive breastfeeding is offering the infant only breastmilk; however, the method of delivery is not described. Enteral feeding is another effective method for providing infants with their recommended fluid intake. Health professionals may consider partial or full enteral feeding due to an infant's stability (fatigue) when feeding or due to airway violation risk in relation to respiratory health. Enteral feeding as management for PFD will be discussed later.

Breastfeeding Versus Bottle Feeding

It is proposed that infants create an intra-oral vacuum during breastfeeding through tongue/jaw movement without peristaltic tongue action and negative pressure for milk ejection (Geddes et al., 2008; McClellan et al., 2010). Conversely, bottle feeding requires repetitive tongue movements in which the base of the tongue moves up and down more than the tip of the tongue, with the tip mainly used to stabilise the teat. An intra-oral vacuum is also created to transport the milk from the teat to the pharynx (Lagarde et al., 2021). Sucking patterns in breastfeeding and bottle feeding differ, with increased suck-per-minute rates found in bottle fed infants (Moral et al., 2010).

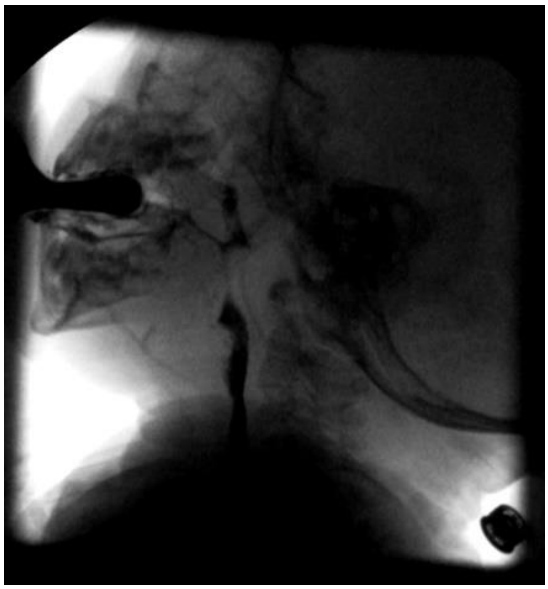
Flow Rate

Bottle and synthetic teat selection is important for supporting and maintaining the infant's physiologic stability while feeding (Goldfield et al., 2006). Bottle systems should be able to maximise volume transfer whilst supporting suck, swallow, breath coordination. Typically, developing infants should be able to control the flow rate within reason by changing the position and movement pattern of the tongue. However, not all infants can do so, and for at-risk populations, the focus may be on external control of flow rate by caregivers. Equipment factors that control flow rate include hole size, teat pliability, teat shape, the position of the fluid

relative to the teat hole, and bottle air exchange (self-pacing bottles) (Ross & Fuhrman, 2015). Infants unable to create suction due to certain craniofacial or neurological disorders require specialised bottle systems that allow for volume transfer solely with compression. These bottle systems may also be beneficial to children with laryngomalacia who have slower total pharyngoesophageal transit times which can be influenced by a greater number of sucks per swallow.

Figure 2

A Videofluoroscopic Image in Lateral View of a 2-Month-Old Child Swallowing Barium, Demonstrating Bottle Feeding



Paced Bottle Feeding

Caregiver responsiveness is also a factor when feeding infants. Breastfeeding does not allow caregivers to control flow rate or fluid volume consumed; however, when compared with bottle feeding caregivers, breastfeeding mothers are more likely to engage in responsive feeding where the caregiver is receptive to infant hunger, stability, and satiation cues (Ventura, 2017). Paced bottle feeding is a method that assists infant stability when bottle feeding(Ventura & Drewelow, 2023). Paced bottle feeding is when the caregiver offers the infant a break every 3–5 sucks by lowering or removing the bottle from the oral cavity, and this has been shown to

support the development of efficient sucking patterns and fewer adverse feeding events (Howe et al., 2007; Thoyre et al., 2012). Paced bottle feeding should especially be considered in children with laryngeal structural abnormalities, such as laryngomalacia. Laryngomalacia causes disordered breathing patterns which may predispose children to altered suck, swallow, breath cycles, adding to risks during feeding.

Positioning

Infants who display physiological instability during bottle feeding might benefit from specific feeding positions to reduce penetration and aspiration risks. These positions include semi-upright position or side-lying position (Park et al., 2014). Breastfed infants have fewer feeding-related oxygen desaturation events when compared to bottle fed infants (Chen et al., 2000; Goldfield et al., 2006). Breastfed infants are usually placed in an unflexed side-lying position, and it has been hypothesised that achieving a similar position may limit adverse bottle feeding events (Dawson et al., 2022). Semi-elevated side-lying has also been reported to support optimal physiological stability during bottle feeds in premature infants (Park et al., 2014). Conversely, Sakalidis et al. (2012) proposed that physiological stability during feeding is due to the intra-oral vacuum theory associated with breastfeeding because physiological stability in bottle fed infants improved when a teat with a vacuum rather than a compression system was used. Positioning in relation to laryngomalacia will be discussed later in this review.

Thickened Fluids

Thickened fluids are a recognised therapeutic intervention that can limit penetration or aspiration events. Thickened fluids increase sucking and oral transit time and reduce PAS scores during VFSS in infants (Gosa, 2012). There are risks when considering thickening feeds for infants, which include reduction in fluid intake; reduced calcium intake; inability to create desired thickness and, in turn, causing continued aspiration with the presence of thickening agent; increased risk of necrotising enterocolitis in infants with a history of prematurity (Gosa et

al., 2011, 2020); and high levels of inorganic arsenic found in infant rice cereals (Bair, 2022). Palatability of feed may change, and the work of swallow is different with thickened feed compared to normal formula or milk viscosity (Gosa, 2012; Gosa et al., 2020). With these risks in mind, thickening should be considered with caution and careful consideration.

Medical Management of Paediatric Feeding Disorders

Children with feeding difficulties due to fatigue, or lack of maturation, presence of aversive feeding behaviours or severe airway violation may require a period of enteral feeding via NGT or percutaneous endoscopic gastrostomy (PEG). Enteral feeding ensures nutritional requirements and growth are met. However, prolonged periods of tube feeding can lead to tube dependency in the absence of continued medical reasons for enteral feeding (Krom et al., 2017). Dependency may be due to delayed development of oral skills (lack of practice) or general refusal and requires a multidisciplinary approach to transition to full oral feeding.

Another aspect of PFD management are specific medical and surgical interventions targeted to address one aspect of dysfunction. As part of holistic care, these may improve PFD outcomes by altering anatomic structures involved in feeding. Surgery may be required for other conditions, not specific to swallowing, and yet may still influence feeding behaviour. The following examples are not an exhaustive list but provide insight into the available surgeries to modify structures involved in different stages of the swallow.

Oral Stage

Tongue reduction surgery for children with macroglossia (protrusion of tongue beyond the level of the alveolar ridge), often associated with Beckwith–Wiedemann syndrome or trisomy 21 is indicated when the tongue causes airway and swallowing difficulties, malocclusion, misalignment of the dental arch and jaw malformation (Kim et al., 2023). Macroglossia typically affects the oral preparatory phase of the swallow and tongue reduction surgery, commonly performed before 2 years old (Simmonds et al., 2018,) has demonstrated

improvement in feeding difficulties and secretion management (Shipster et al., 2012). Surgery improves mandibular prognathism (Kim et al., 2023), which supports oro-motor development and skills required for solid foods and improved airway patency is hypothesised to support swallowing outcomes. Removal of obstructive tonsils (tonsillectomy) may increase oropharyngeal space and prevent food impactions or aversive behaviours, encouraging oral intake.

Pharyngeal Stage

Surgical repair (endoscopic surgery or injection laryngoplasty) for laryngeal clefts may be required for children who do not respond to conservative treatment as evidenced by persistent aspiration, recurrent respiratory symptoms and faltering growth (Timashpolsky et al., 2021). In a systematic review and meta-analysis, Timashpolsky et al. (2021) reported that the resolution of aspiration events ranged from 50–71% following surgical repair. Feeding implications associated with supraglottoplasty surgery for children with laryngomalacia will be discussed later in the review.

Oesophageal Stage and Reflux

The international consensus for the definition of GERD is the “passage of gastric contents into the oesophagus with OR without regurgitation and/or vomiting” (Rosen et al., 2018, p. 546). Reflux disease (GERD) occurs when this escape causes tissue damage or symptoms (Montreal classification). Reflux is common in infancy due to the relatively large liquid diet and horizontal positioning with 70–85% of infants displaying symptoms of reflux in the first 2 months of life (Vandenplas, 2014). GERD is often self-limiting and resolves with the introduction of solid foods and increasingly upright developmental positioning. Conversely, GERD may cause “troublesome symptoms that affect daily life”, and symptoms of GERD in infancy may include crying, back arching, regurgitation, and irritability (Rosen et al., 2018). GERD is often present in children who have comorbidities such as prematurity, neurological

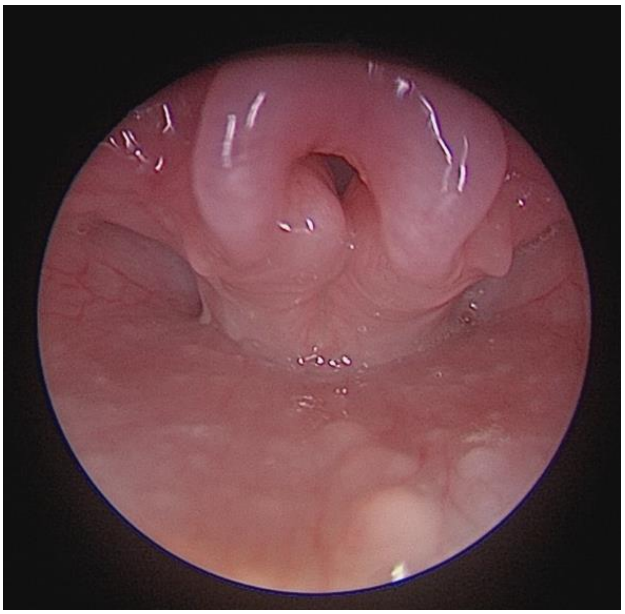
impairment, respiratory complications, and congenital anomalies (Rosen et al., 2018). GERD also increases the risk of childhood feeding disorders due to a range of problems (Sdravou et al., 2019), including appetite suppression, faltering growth, recurrent aspiration pneumonia (Gulati & Jadcherla, 2019) and in extreme cases, cardiorespiratory failure (Rosen et al., 2018). In 2018, the *Paediatric Gastroesophageal Reflux Clinical Practice Guidelines* for North America and Europe were published. The guidelines for children under 12 months of age recommend taking a thorough medical and feeding case history and additional diagnostic testing as required. Intervention is a stepwise approach beginning with limiting fluid volumes, thickening feeds and breastfeeding, followed by specialist formula/ cow's milk protein allergy (CMPA) testing, and finally, acid suppression/medication (Rosen et al., 2018). However, acid suppression therapy in infancy should be used with caution as reflux in infants is typically nonacidic (Hartl & Chadha, 2012), which may impact the effectiveness of acid suppression (Duncan et al., 2021). Acid suppression therapy has also been found to increase the rate of respiratory infections in infants with laryngomalacia (Duncan et al., 2021). Anti-reflux surgery, namely fundoplication, is indicated specifically in chronic conditions with life threatening GERD complications when all other methods to manage GERD and GERD-related complications have failed. Fundoplication in the paediatric population improved GERD symptoms by 86% and postoperative dysphagia occurs less frequently after partial fundoplication (Mauritz et al., 2011). Risks of surgery include bloating, early satiety, pain, impaired bolus transfer, aspiration related to oesophageal stasis, retching, dumping syndrome, and the need for repeat surgery (Rosen et al., 2018).

Chapter 2 Laryngomalacia

Laryngomalacia is a congenital anomaly of the larynx found in infants and is characterised by inward prolapse of flaccid supraglottic structures during inspiration (see Figure 3) (Bedwell & Zalzal, 2016; Hartl & Chadha, 2012). Abnormalities that may be present include shortened aryepiglottic folds that collapse medially, an “omega-shaped” or retroflexed epiglottis collapsing into the laryngeal vestibule on inspiration, and prolapsing of redundant mucosa that overlays the arytenoid cartilages (Gan et al., 2021; Klinginsmith et al., 2019; Olney et al., 1999).

Figure 3

Endoscopic Images of the Larynx During Inspiration Demonstrating Concentric Constriction of the Laryngeal Vestibule. With Thanks to Auckland Hospital



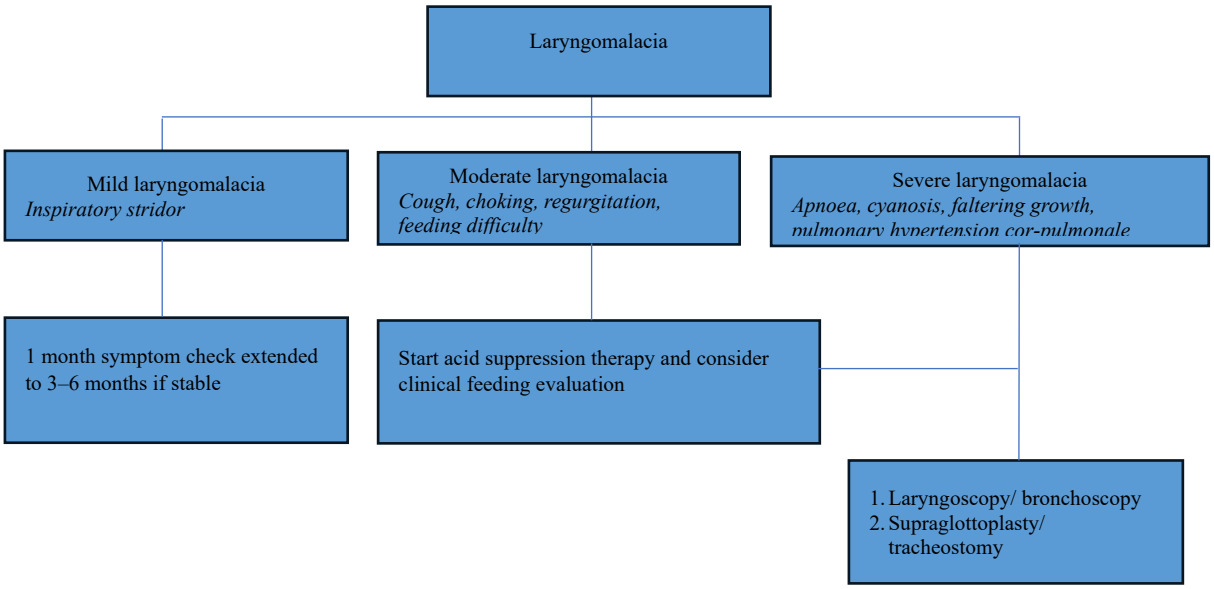
Several laryngomalacia classification schemes have been proposed (Sidell & Messner, 2021). Earlier publications largely use the Holinger scale, whereas the Olney classification system is used in later research. The Olney classification scale demonstrates stronger inter-rater reliability scores than the Holinger (Sivarajah et al., 2020). The Olney classification scale used by paediatric otolaryngologists classifies laryngomalacia into three subcategories based on the site of supraglottic obstruction. Type 1 is the prolapse of mucosa overlying the arytenoid

cartilages, type 2 is foreshortened aryepiglottic folds, and type 3 is the posterior displacement of the epiglottis (Olney et al., 1999).

In addition to previously published classification systems, Carter et al. (2016) introduced a symptom and severity based classification scheme that can be used to determine the intervention approach (see Figure 4). *The International Paediatric ORL Group (IPOG) Laryngomalacia Consensus Recommendations* provide clinicians with a stepwise algorithm for treatment, including treatment options and when to refer to a specialist.

Figure 4

. Laryngomalacia Intervention Approach. Adapted From Carter et al. (2016)



Prevalence

Laryngomalacia is the most common laryngeal anomaly and cause of chronic inspiratory stridor in neonates (Bedwell & Zalzal, 2016; Carrion et al., 2018; Gan et al., 2021; Zoumalan et al., 2007). The exact incidence of laryngomalacia is unknown and underreported as many infants are managed conservatively, with estimates ranging between 1 in 2,000 to 1 in 3,000 (Klinginsmith et al., 2019). Approximately 99% of infants with laryngomalacia are categorised as mild or moderate (Carter et al., 2016; Green et al., 1983). Premature infants as well as

Hispanic and Black infants of all gestational ages are at a higher risk of this laryngeal anomaly (Edmondson et al., 2011), with a general male predominance (1.9:1) (Cooper et al., 2014).

Onset and Resolution

Laryngomalacia symptom onset is typically within the first few weeks of life (Bedwell & Zalzal, 2016), and symptoms can be expected to peak between 6 and 8 months (Thompson, 2007), with improvement by 12 months and resolution by 18–24 months (Carrion et al., 2018). However, Isaac et al. (2016) argue that there is limited evidence available regarding time and rates of resolution, and a study by Hilland et al. (2016) concluded that whilst cases are typically self-limiting, laryngomalacia leaves structural and functional footprints in the larynx that increase risk of exercise-induced symptoms and laryngeal obstruction well into adolescence. Therefore, due to the purported differing times to resolution, feeding problems in this population can be characterised as either acute or chronic, and early identification may improve feeding outcomes.

Pathophysiology

There is still no consensus regarding the pathophysiological mechanism involved in laryngomalacia (Gan et al., 2021). However, two proposed recent theories are *chondropathic* (abnormal cartilaginous development) or *neurological dysfunction* due to neuromuscular hypotonia (Bedwell & Zalzal, 2016; Gan et al., 2021). A study by Gan et al. (2021) found that 70.5% of children who had undergone aryepiglottoplasty had signs of inflammation of the laryngeal mucosa, implying a mild concurrent laryngitis/supraglottitis in most cases. The authors also discussed the presence of immature cartilage, which supports the theory of a chondropathic element in the aetiology.

Conversely, sensorimotor integrative function has also been shown to be reduced in this population (Klinginsmith et al., 2019). Inflammation of the mucosa could initiate functional denervation and blunting of afferent reflexes (Gan et al., 2021), which would explain the

decreased laryngeal sensation and possibly infers a neurological component (Thompson, 2007). The cranial nerves involved in swallowing have both sensory and motor components, with the exception of cranial nerve XII (hypoglossal nerve). It is hypothesised that sensory (laryngopharyngeal sensation) and motor deficits are present (Arvedson & Lefton-Greif, 2017) and contribute to swallowing deficits in laryngomalacia. A study by Munson et al. (2011) further supports the neurologic dysfunction theory, as nerve hypertrophy was found in supra-aryepiglottoplasty specimens of children with laryngomalacia when compared with tissue from controls. The sensorimotor integration of the laryngeal adductor reflex (LAR) is thought to be impaired, impacting laryngeal tone and leading to secondary consequences of poor pharyngeal secretion management, airway violation, and apnoeic episodes (Thompson, 2007).

Interestingly, decreased laryngeal sensation is also found in GERD, and there is an increased prevalence of GERD in children with laryngomalacia, which also correlates with laryngomalacia severity (Hartl & Chadha, 2012). Fifty-nine percent of infants with laryngomalacia have reflux which is significantly higher than the general infant population, where estimates range from 2.5–33% (El-Serag et al., 2014). It is important to consider neurological comorbidity as children with neurodisabilities also have an increased incidence of GERD (Trinick et al., 2012). Hysinger (2018) argued that, theoretically, a resistant airway could cause worsening reflux with laryngeal oedema, exacerbating airway obstruction in a cyclical process. There is no direct evidence of a causal association between reflux and laryngomalacia, but reflux may worsen upper airway obstruction through irritation and oedema (Klinginsmith et al., 2019).

Infants with laryngomalacia often have additional comorbidities, including neurologic disease, cardiopulmonary disease, congenital anomalies, neonatal abstinence syndrome and other syndromes (Abraham et al., 2022; Thompson, 2007). Up to 20% of infant laryngomalacia cases present with neurological conditions supporting the theory of neurological involvement

(Thompson, 2007). However, whilst laryngomalacia is typically congenital, acquired laryngomalacia is also possible following damage to the central nervous system, with the insult occurring at the brainstem nuclei for airway patency causing abnormal integration of sensorimotor function (Thompson, 2007). Acquired laryngomalacia can occur in both adults and children (i.e., stroke, seizures, hypoxic injury, sedation) and the resolution of symptoms is observed following resolution of the neurological condition (Thompson, 2007). Thompson (2007) therefore also proposed that congenital laryngomalacia is a consequence of impaired or immature sensorimotor brainstem reflexes. It is believed that neurological conditions influence both severity and outcomes of supraglottoplasty as approximately 8% of patients with mild laryngomalacia also have neurological conditions, rising to 11% in those with moderate disease and 34% for severe disease (Klinginsmith et al., 2019). Thus, the prevalence of neurologic conditions in laryngomalacia supports the theory that a causative neurological element exists.

Diagnosis

The diagnosis of laryngomalacia requires visualisation of the larynx using flexible nasopharyngoscopy. This allows the ORL surgeon to rule out other airway pathologies such as subglottic stenosis, vocal fold paralysis, and GERD (Jacobs, 2022; Moroco & Aaronson, 2022). However, in cases of mild laryngomalacia, children are often diagnosed by signs and symptoms alone and clinically followed by community developmental paediatricians and not diagnosed endoscopically (Klinginsmith et al., 2019). There is also evidence to suggest that patients with identified laryngomalacia should undergo a microlaryngobronchoscopy (MLB) because there is an increased prevalence of synchronous airway lesions (SALs) associated with more severe clinical presentation as well as neurodevelopmental conditions causing additional morbidity risk (Glibbery et al., 2022).

Symptoms

Infants with laryngomalacia often present with a range of signs and symptoms, including apnoea, respiratory distress, feeding problems, and faltering growth as a result of increased work of breathing and PFD (Bedwell & Zalzal, 2016). The most common type of laryngomalacia is Olney type 1 which predominates in children presenting with snoring or sleep disordered breathing (Cooper et al., 2014). Infants with airway compromise typically have difficulties feeding (Jadcherla, 2017), and in a retrospective case series, 86% of children with laryngomalacia presented with swallowing difficulties (Scott et al., 2019). Anecdotally, stridor worsens during feeding or agitation, which is correlated with positioning, increasing when the infant is placed in supine (Bedwell & Zalzal, 2016; Carrion et al., 2018). However, the consequences of agitation are not universal; crying typically improves stridor in infants with mild to moderate laryngomalacia due to improved motor tone and worsens in infants with severe laryngomalacia due to an increase in airflow (Sidell & Messner, 2021).

Medical Management of Laryngomalacia

Patients may be managed either conservatively or surgically or a combination of both, and a holistic approach should be considered due to the heterogeneity of children with laryngomalacia (Carter et al., 2016).

Non-surgical approaches including symptom review, acid suppression therapy for reflux management, and feeding therapy, are considered appropriate for those with mild to moderate laryngomalacia (Carter et al., 2016).

Surgical options for improving symptoms of laryngomalacia include *supraglottoplasty*, a procedure that divides the aryepiglottic folds and removes redundant arytenoid tissue or *tracheostomy*, a procedure where a communication (stoma) is made from the trachea to skin surface to create a patent airway. When considering surgical intervention, issues relating to respiratory distress, suboptimal feeding, and weight gain are cited as indications for treatment,

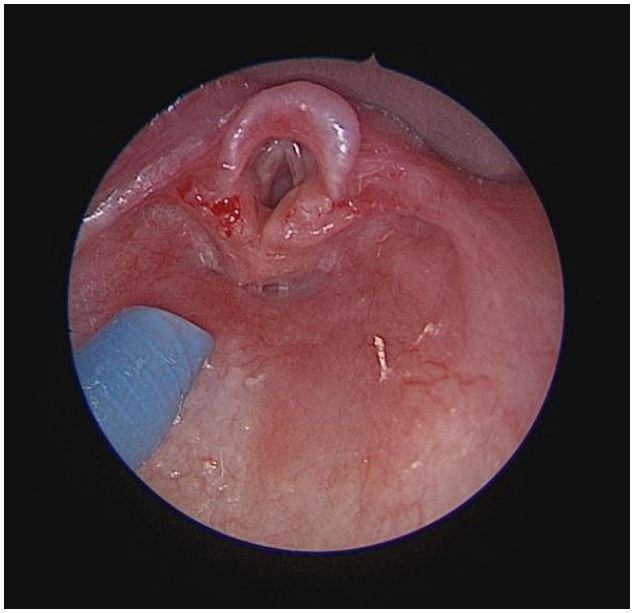
and all severe cases should be considered for surgery (Jacobs, 2022). Overall, 10% of patients require surgical treatment and approximately 25% of children who initially present with moderate symptoms will later require surgery (Bedwell & Zalzal, 2016; Sidell & Messner, 2021).

Historically, tracheostomy was performed to create a sustained patent airway (Holinger & Konior, 1989); however, supraglottoplasty is the current surgical preference in most cases (Bedwell & Zalzal, 2016). Supraglottoplasty is typically performed with surgical instruments either steel instruments (cold supraglottoplasty) or with a carbon dioxide laser (Jacobs, 2022; Sidell & Messner, 2021). Surgery is usually performed between 3 and 5 months of age (Bedwell & Zalzal, 2016), although time frames differ globally with the procedure in a regional European service performed between 3 weeks and 36 months of age (Gan et al., 2021).

Risks involved with supraglottoplasty include postoperative supraglottic stenosis (Jacobs, 2022), incomplete response, damage to the vocal folds, or worsened postoperative aspiration. According to the *IPOG Laryngomalacia Consensus Recommendations*, providers must consider comorbidities that may lead to suboptimal outcomes when deciding surgical treatment options. For example, supraglottoplasty is contraindicated in children with multiple and severe comorbidities, including those with an unsafe swallow associated with a neurological condition such as cerebral palsy or generalised pharyngo-laryngomalacia with pharyngeal collapse (Carter et al., 2016; Jacobs, 2022; Simons et al., 2016). Therefore, children with neurologic or cardiac conditions typically experience significantly higher tracheostomy rates (Hoff et al., 2010).

Figure 5

Endoscopic Images of the Larynx During Inspiration Demonstrating Typical Supraglottic Structures Post Supraglottoplasty



Paediatric Feeding Disorders and Laryngomalacia

Swallowing dysfunction is reported in the laryngomalacia population with several studies attempting to estimate the true prevalence and presence of aspiration. To date, studies have not described the biomechanics leading to airway violation or bolus retention in this cohort of children. Estimates of children with laryngomalacia who experience aspiration range from 49- 90.1%; however, there is limited evidence of a correlation between the severity of laryngomalacia and the presence of oropharyngeal dysphagia (Irace et al., 2019; Jaffal et al., 2020; Scott et al., 2019; Simons et al., 2016). It is also important to note that common comorbidities seen in laryngomalacia are also associated with PFD, and it is difficult to account for the effect of these variables when examining the prevalence and causation of oropharyngeal dysphagia in laryngomalacia. Nevertheless, studies suggest that this laryngeal anomaly alone may cause incoordination in the suck swallow breathe pattern required for infant feeding (Jaffal et al., 2020; Simons et al., 2016).

Children with laryngomalacia frequently have identified airway violations during subjective and instrumental assessment (Irace et al., 2019; Simons et al., 2016). In a large retrospective cohort study of 324 children, with and without comorbidities, at a tertiary centre who underwent swallowing assessment, “dysphagia” or “feeding difficulty” was noted in 163 children (50.3%) (Simons et al., 2016). Three different assessment protocols were used, namely, clinical feeding evaluations in 53 children (16.3%), VFSS in 72 children (22.2%) and FEES in 130 children (40.1%). No significant relationship was found between the severity of laryngomalacia, comorbidities (i.e. syndromes or GERD) and penetration or aspiration. However, there was an increased likelihood of faltering growth associated with greater severity of laryngomalacia. There may be a sampling bias in this study, as 62.7% of children presented with mild laryngomalacia which was treated conservatively by a developmental paediatrician (Klinginsmith et al., 2019) and children were recruited for swallowing assessments mainly due to dysphagia symptoms which may have occurred only in the more severe cases. There are additional issues in this study, such as a non-blinding bias and the use of different methods (clinical feeding evaluation, VFSS and FEES) for detecting swallow dysfunction. Detecting the true prevalence of dysphagia in children with laryngomalacia may not be possible from this study.

In a similar retrospective review of 142 children with laryngomalacia with and without comorbidities who underwent VFSS, 128 children (90.1%) had identified swallowing difficulties and 60 (42.4%) aspirated, with 59 doing so silently (59/60, 98.3%) (Irace et al., 2019). Forty children (28.2%) had identified penetration (PAS 2–5) without aspiration. Unlike Simons et al.’s (2016) findings, comorbidities including seizure disorders, laryngeal cleft, and prematurity increased the risk of airway violation during VFSS. However, the findings should again be interpreted with caution when attempting to extrapolate the prevalence of penetration or aspiration in the wider laryngomalacia population. All children recruited to this study either

had reported respiratory issues and/ or feeding difficulties and thus whilst penetration and aspiration, specifically silent aspiration was highly prevalent in this study, clinical symptoms at bedside were present in order to receive a VFSS. This is a selection bias and cannot represent the true prevalence of airway violation risk in the broader pool of children with laryngomalacia. As with most retrospective studies evaluating quantitative swallowing assessments, the authors were not blinded, and false positives may have occurred. Furthermore, the study only identified "swallowing dysfunction" through documented penetration or aspiration (PAS 2–8) during VFSS without accounting for other known risks such as residue. The authors also commented that VFSS captures the swallow at one point in time and penetration or aspiration events may have been missed.

What is clear from both studies is that clinicians should consider airway violation in infants with laryngomalacia who present with recurrent respiratory issues. The question of the true prevalence of silent aspiration in this population should be examined further, as both studies are based on symptomatic children receiving swallowing assessments due to subjective swallowing concerns or recurrent respiratory issues, introducing selection bias.

Laryngomalacia, Surgery, and Paediatric Feeding Disorders

Surgery is not without complications. Reported complications following supraglottoplasty include increasing the risk of postoperative aspiration, although this is typically temporary (Anderson de Moreno et al., 2015; Rastatter et al., 2010; Schroeder et al., 2008) and is contested somewhat in the literature (Richter et al., 2009). A small retrospective cohort study of 44 children, with and without comorbidities, including genetic syndromes or neuromuscular disorders reported that 92% of children had "improved dysphagia", which included less restrictive diets and "improved" follow-up VFSS postoperatively; however, the study was unable to calculate statistically significant change in penetration or aspiration scores due to limited follow-up data (Scott et al., 2019). The study also reported that children with

underlying genetic syndromes were more likely to have persistent swallow dysfunction postoperatively. The study did not publish specific data regarding the presence of airway violation events at baseline or follow-up, making comparison with other studies difficult. In another small but contrasting retrospective study, Rastatter et al. (2010) examined 39 patients with severe congenital laryngomalacia without neurological comorbidities who underwent supraglottoplasty. Despite silent aspiration concerns in this population that it is not always detected during a clinical feeding examination, similar to the study by Simons et al. (2016), a clinical feeding examination or VFSS determined the presence of aspiration. A clinical feeding examination alone detected aspiration in 69.2% of cases preoperatively and 20.5% of cases postoperatively. Ten children (25.6%) aspirated preoperatively, and two of these children (20%) improved 48 hours postoperatively. However, this study also found new onset penetration and aspiration in 13/29 children postoperatively (44.8%). It is important to note that aspiration is a well-documented risk in laryngomalacia regardless of surgical intervention and aspiration in infants with multiple comorbidities may not improve postoperatively (Jadcherla, 2017; Moroco & Aaronson, 2022; Schroeder et al., 2008).

Irrespective of the limited and conflicting results surrounding supraglottoplasty and swallowing outcomes, there is a perception of improved feeding outcomes postoperatively by parents. Kanotra et al. (2018) completed a parental perception of swallowing questionnaire in 28 parents pre and post supraglottoplasty. They found a significant improvement in the overall parental perception of swallowing in children with laryngomalacia, including fewer “choking” episodes, fewer breathing issues associated with feeding, less overt reflux and increased oral intake. However, the questionnaire was not a standardised assessment tool, questionnaires were completed 3 months post-surgery and surgeries were completed by one surgeon, and potential developmental, spontaneous improvement was not considered. Nevertheless, parents of children with laryngomalacia who had airway violation issues confirmed through FEES have reported

significant emotional impact and perceived worsening infant health for children requiring supraglottoplasty on a validated genetic parental questionnaire (Thottam et al., 2016). This highlights that parents of children with laryngomalacia who require surgery have substantial stress related to the disease, and in the absence of a clear consensus regarding the relationship between swallowing outcomes and surgery, there is perhaps a placebo effect, power of expectation (Brown, 2013) or recall bias present. Regardless, as Kanotra et al. (2018) stated that caregiver satisfaction regarding feeding will ultimately influence the perceived impact of surgery.

Interventions for Paediatric Feeding Disorders in Laryngomalacia

A number of considerations regarding intervention specifically for children with laryngomalacia must be acknowledged prior to intervention commencing.

Enteral Feeding Considerations

NGT placement following supraglottoplasty may cause inflammation and recurrent stridor or obstruction (Sidell & Messner, 2021). Therefore, other long term enteral feeding options, such as a gastrostomy, may be considered when aspiration cannot be mitigated through feeding interventions such as thickened fluids or controlling flow rate. However, in most cases penetration and aspiration postoperatively are thought to be self-limiting. In a retrospective cohort study, Schroeder et al. (2008) found that in all laryngomalacia cases with and without comorbidities, postoperative aspiration (89% of children) quickly resolved and children who required long term enteral feeding (gastrostomy) had concurrent neurological conditions with 4/7 children requiring continued enteral feeding at 2 years follow-up.

Reflux Management and Thickened Feeds

The *IPOG Laryngomalacia Consensus Recommendations* include feeding therapy and acid suppression in improving laryngomalacia outcomes. A retrospective cohort study of 236 children assessed the role of acid suppression or thickened feeds in laryngomalacia outcomes

(Duncan et al., 2021). The authors found that children treated with acid suppression were more likely to require early supraglottoplasty. Children treated with acid suppression were also twice as likely to be hospitalised due to risks associated with respiratory illness when compared to those treated with thickened fluids. Interestingly, children with swallow dysfunction identified through a clinical feeding evaluation (27% overt signs of aspiration) or VFSS (69% penetration or aspiration) were not more likely to receive surgical intervention ($p = .202$). Reflux was attributed to feeding difficulties in 13% of children in the swallowing evaluation group who were more likely to be offered acid suppression. A clinical feeding evaluation and VFSS were completed to identify aspiration; agreement between the two assessment methods was evaluated, demonstrating a significant difference between the two methods. A normal bedside assessment was reported in 29/36 children who were subsequently identified as having an abnormal follow-up VFSS. Seventy-one percent of children demonstrated silent aspiration in this study and the poor agreement between the two assessment methods implies that the MDT should consider instrumental swallow assessments to diagnose and manage airway violation in children with laryngomalacia. However, caution must be used with these findings due to the retrospective nature of this study and that only children with more severe symptoms were treated, limiting the representativeness of the wider laryngomalacia population.

Whilst thickened fluids in this population may treat GERD and decrease laryngeal penetration, thickening feeds without VFSS should still be considered with caution (Duncan et al., 2019, 2021). In a retrospective cohort study of 137 children with laryngomalacia who showed laryngeal penetration without aspiration on VFSS, Duncan et al. (2019) found that following feeding intervention (thickened fluids or flow rate change), 77% of children had symptom improvement when compared to the no intervention group (16%) and those in the thickened fluids group had the greatest improvement on VFSS (91%), which was significant. However, as previously discussed, not all penetration or aspiration events are able to be captured

during VFSS and 26% of children demonstrated penetration or aspiration during their follow-up VFSS that was not captured in the initial study, highlighting the issues associated with this quantitative swallow assessment. Furthermore, symptom improvement was subjective based on caregiver report which may be open to bias. The study did not explore underlying biomechanics in the children or how thickened fluids or a change in flow rate impacts swallow pathophysiology in children with laryngomalacia, as quantitative metrics were not utilized (Duncan et al., 2019).

Positioning

Positioning infants in prone to decrease stridor is widely reported (Van Heest et al., 2018). Optimal feeding positions may also be important in limiting airway violation events. Placing infants with laryngomalacia in semi-prone to limit aspiration risk during breastfeeding has been found to be effective as it reduces dynamic supraglottic soft tissue collapse, increases the volume capacity of the pyriform fossae, and reduces the flow rate of milk (Mills et al., 2021). Mills et al. (2021) used FEES to assess infant swallowing (n = 23). This study found signs of dynamic airway obstruction or impaired airway protection when swallowing in 87% of infants when placed in a supine or semi lateral position; however, when these 20 infants were repositioned to semi prone, improvement or resolution of stridor and latch was observed in all infants. The authors acknowledge the results are subjective but argue the use of FEES in this population may be more appropriate because it allows observation of both the abnormal pharyngeal and laryngeal anatomy. To date, studies have not focused on positioning or paced bottle feeding in this population during VFSS or FEES.

Literature Gap

Most studies in the current literature are retrospective and observational, often medical chart reviews. There is a paucity of published randomised controlled trials or blinded studies in children with laryngomalacia using either a VFSS or FEES to assess the impact of feeding

therapy strategies or surgery on swallow biomechanics. This is understandable due to the ethical issues in withholding treatment and requiring caregiver consent for treatment. However, the generalisation of results is limited due to the retrospective designs and tertiary-centre nature of most studies. There is still a need for prospective longitudinal studies that identify causations and not correlations in cumulative (specific comorbidities) and mitigating factors (surgery/feeding therapy strategies) associated with penetration and aspiration risk as well as identifying the true prevalence and underlying cause of airway violation events in this population (Duncan et al., 2019; Irace et al., 2019). The introduction of standardised swallow assessment for all studies would support future consistency in reporting of findings.

Chapter 3 Aims and Purpose of This Study

Rationale

There is a growing knowledge base regarding PFD and laryngomalacia. However, there is still a lack of research studies focusing on swallowing biomechanics in children with laryngomalacia using paediatric VFSS quantitative measures, while also analysing longitudinal swallow outcomes. There is also variability in the literature regarding the prevalence of penetration and aspiration, and postoperative aspiration rates in this population. Introducing a quantitative and objective VFSS assessment may support future studies in developing comparable data and perhaps reducing variability in findings.

Aim

The overarching aim of this study was to describe the swallowing biomechanics of bottle fed children with laryngomalacia using published quantitative videofluoroscopic swallow measures and retrospective VFSS recordings. This study also aimed to identify which quantitative measures in children with laryngomalacia, with and without coexisting comorbidities, are associated with increased risk of airway violation. This study hopes to provide preliminary evidence to support the implementation of standardised swallowing assessment protocols for children with laryngomalacia both pre- and post-operatively.

Research Questions

1. Are there changes in timing or displacement fluoroscopic measures that increase the risk of penetration or aspiration in bottle fed children with laryngomalacia?
2. Does penetration and aspiration in children with laryngomalacia improve over time, and what are the swallow biomechanical risk factors associated with continued penetration and aspiration?

3. Are the swallow biomechanics similar in children with laryngomalacia in isolation, compared to children with laryngomalacia and comorbidities?

Hypotheses

1. There will be a strong correlation between quantitative timing measures on VFSS and penetration or aspiration measures (PAS scores).

2. The severity of penetration and aspiration will decrease as children get older in accordance with general disease improvement and improvement of other laryngomalacia symptoms.

3. Swallow biomechanics, specifically quantitative airway timing measures, will be similar in all children with laryngomalacia; however, displacement measures (such as pharyngeal constriction) and bolus retention/ redirection may be more prevalent in children with a concurrent chromosomal or neurological component.

Significance

Analysing the swallow biomechanics in children with laryngomalacia will add to the understanding of how swallowing is impaired. This will provide the multidisciplinary team with a greater ability to interpret VFSS findings and choose directed interventions. It may also provide an understanding of the relative (predictive) risk of penetration and aspiration events occurring in bottle fed children even when penetration or aspiration has not been captured during the VFSS assessment. A quantitative assessment protocol with an understanding of underlying biomechanical impairment will support decisions and timing of optimal surgical and non-surgical interventions for these children.

Proposed Study

The study will build on a series of papers from The University of Auckland Swallowing Research Laboratory (Dharmarathna et al., 2021; Fuller et al., 2022; Miles et al., 2022), which

explored the reliability and clinical relevance of quantitative videofluoroscopic measures in the paediatric population. This retrospective longitudinal observational cohort study explores VFSS studies of 20 children who received a laryngomalacia diagnosis via direct laryngoscopy or laryngobronchoscopy presenting with bottle feeding difficulties.

Chapter 4 Methods

This single centre retrospective longitudinal observational cohort study included 26 children who received a laryngomalacia diagnosis via direct laryngoscopy or laryngobronchoscopy and presented with bottle feeding difficulties who underwent at least one fluoroscopic study at one tertiary children's hospital from 2012–2022. A retrospective clinical record audit at this tertiary hospital found 877 children with a diagnostic code for laryngomalacia, with 228 (26%) seen by speech pathology and 26 (3%) receiving a VFSS. Children were recruited through The University of Auckland Swallowing Research Laboratory's videofluoroscopy database of children consecutively referred for VFSS for feeding difficulties. Upon further analysis of medical records, six children were excluded due to: a) noncompliance on VFSS (n = 1); b) VFSS image quality prevented the reliable collection of data (n = 1); c) non-bottle fed during VFSS (n = 2); d) wrongly coded as laryngomalacia in medical records (n = 2). Twenty children remained in the study for analysis. The study received ethical approval from The University of Auckland Human Participants Ethics Committee (application number: 9263).

Video Fluoroscopic Swallow Study Procedure

VFSS were conducted in the radiology suite at a tertiary children's hospital according to a standardized protocol (Henderson et al., 2016) using a Siemens Sireskop radiographic unit (Siemens, Munich, Germany) in lateral view. Studies were initially captured using continuous fluoroscopy recorded at 25 frames per second directly onto the hospital picture archiving and communication system (PACS). Videos were subsequently exported to the University of Auckland Swallowing Research Laboratory's videofluoroscopy database for children. All studies were performed by a speech pathologist and radiologist. All children were offered International Dysphagia Diet Standardisation Initiative (IDDSI, 2019) Varibar® Level 0 Thin Liquid barium sulfate powder for suspension (40% w/v) (E-Z-EM Canada Inc, Quebec, Canada) through a bottle and positioned in an upright supportive seating system with the support of a

caregiver or speech pathologist. Due to the retrospective nature of this study, it was not possible to control for bottle brand, teat size, bolus size or suck feed timing and all studies were clinically led at the speech pathologist's direction.

A total of 23 studies and 156 swallows were analysed; the number of swallows per study ranged from 2–12. The total number of analysed swallows from children's first VFSS was 147 (M = 7 swallows per child). Seven children underwent repeat VFSS (35%). Repeat VFSS were excluded if the child was offered a different method of fluid intake (i.e., cup drinking). Therefore, four VFSS studies were excluded from the study due to cup or spoon feeding, leaving three repeat bottle fed VFSS studies (total nine swallows) for follow-up analysis.

Video Fluoroscopic Swallow Study Analysis

Data collection and analysis were performed from March 2023 to February 2024 by the author. This included a review of the children's medical records, collecting demographic information, operative/clinical reports, and VFSS findings. All VFSS were analysed using the software program, Swallowtail, which allowed for frame-by-frame quantitative and objective analysis of VFSS based on Leonard and Kendall's research (Swallowtail™, Belldev Medical, Illinois, USA). The author completed both face-to-face training and online recorded training on Swallowtail measures based on the Lab's published VFSS protocol (Miles et al., 2022).

Table 3 provides an in-depth description of all Swallowtail measures used. Only two studies included the use of a radiopaque ring to allow for recording additional displacement measures; therefore, the maximum opening of the pharyngoesophageal segment (PESmax) was not recorded or analysed. Hyoid excursion was also not recorded as the hyoid is difficult to visualise in infants under 9 months of age (Riley et al., 2019). Timing and displacement measures were collected and calculated using built-in tools within the Swallowtail program, and manually entered into an Excel spreadsheet for analysis (see Table 3).

Table 3

Objective and/or Quantitative Swallow Measures of Children

Objective measure	Definition
Timing(s)/Coordination (Leonard & Kendall, 2019; Miles et al., 2022)	
Total pharyngeal transit time (TPT)	Represents the total time of the bolus passage through the pharynx, from when the bolus head passes the posterior nasal spine, to the time at which the bolus tail completely clears the PES.
Time to airway closure (Airwaycl)	Time taken to total arytenoid- epiglottis approximation indicating total supraglottic airway closure.
PES opening duration (PESdur)	The duration of PES opening from the first frame in which it opens, to when it closes behind the bolus tail.
Coordination of airway closure with bolus transit (BP1AEcl)	Airway closure time in relation to bolus reaching the PES.
Number of sucks per swallow	On suck is defined as the downward motion of mandible- to-mandible returning to neutral position. This measure assesses the total number of sucks per swallow.
Displacement measures (cm) (Miles et al., 2022)	
Pharyngeal constriction ratio (PCR)	The ratio of pharyngeal area at maximum constriction to the area of the pharynx at rest.
Bolus Clearance Ratio (BCR)	The ration of residue present in the pharynx after PES closure and relaxation of the pharynx.
Descriptive swallow measures	
Penetration- aspiration scale (Rosenbek et al., 1996)	Objective scale to identify penetration and aspiration. Please refer to Table 4.
Nasopharyngeal redirection (NPR)	Presence or absence of NPR.
Pharyngoesophageal redirection	Presence or absence of pharyngoesophageal redirection.
Residue	Presence of absence of residue in the pharynx.

Signs of Airway Violation and Residue Measures

Signs of airway violation (penetration and aspiration), bolus retention and redirection, were also recorded. Penetration and aspiration for each individual swallow was recorded using PAS (Rosenbek et al., 1996). A PAS score of 3 or higher was recorded as an incident of airway violation (Daggett et al., 2006; Dharmarathna et al., 2021; Steele et al., 2017) (see Table 3). Maximum PAS score was also recorded to understand the severity of the airway violation events for each study. Residue, nasopharyngeal, and pharyngoesophageal redirection were recorded as

binary events (present or absent) for each swallow. Due to the distinctive suck swallow breathe pattern in bottle-feeding infants (Arvedson & Brodsky, 2002), the number of sucks per swallow was recorded.

Table 4

Penetration-Aspiration Scale (Rosenbek et al., 1996)

Score/Classification	Description
1 None	Material does not enter the airway
2 Penetration	Material enters the airway, remains above the vocal folds, and is ejected from the airway
3 Penetration	Material enters the airway, remains above the vocal folds, and is not ejected from the airway
4 Penetration	Material enters the airway, contacts the vocal folds, and is ejected from the airway
5 Penetration	Material enters the airway, contacts the vocal folds, and is not ejected from the airway
6 Aspiration	Material enters the airway, passes below the vocal folds, and is ejected into the larynx or out of the airway
7 Aspiration	Material enters the airway, passes below the vocal folds, and is not ejected from the trachea despite effort
8 Aspiration	Material enters the airway, passes below the vocal folds, and no effort is made to eject

Note. PAS 3–8 = Airway violation.

Video Fluoroscopic Swallow Study Reliability

Analysis of VFSS data was completed by the author and 31 individual swallows (20% of the total number of swallows) were randomly selected for inter-rater reliability rating and evaluated by an experienced speech pathologist. The author and speech pathologist reviewed all 31 individual swallows together, and where disagreement between measures occurred, the measure was reviewed, and consensus was obtained.

Data Analysis

Descriptive statistics were collated using Excel (Microsoft, Seattle, USA) and transferred to IBM SPSS Statistics version 24 (SPSS Inc., Chicago, Illinois, USA) for further inferential

analysis. The median, mean and range values from the total number of swallows for each child's study were calculated and entered into a spreadsheet. As the sample size was small and quantitative measures were not normally distributed, non-parametric tests were completed in order to assess statistical significance with the median representing the centre of distribution. All statistical tests were two-sided with *p*-values > .05 considered statistically significant. Spearman's correlations and chi-squared tests were performed to determine associations between swallow and demographic/clinical measures. Box plots were also created for measures that had statistically significant correlations in order to visually explore relationships.

The cohort was categorised into three diagnostic subgroups in order to analyse and compare the clinical background, quantitative and symptom measures between isolated laryngomalacia and laryngomalacia with comorbidities. These groups were: 1) laryngomalacia in isolation, 2) laryngomalacia with a concurrent chromosomal or neurological component, and 3) laryngomalacia with a concurrent additional anatomical abnormality. Kruskal–Wallis one-way ANOVA and Mann–Whitney tests were used to analyse differences in swallow measures across groups.

The values for each child (within each subgroup) were compared to reference values that summarised from the previously published mixed aetiology VFSS data by The University of Auckland Swallowing Research Laboratory (see Table 5).

Table 5

Quantitative and Objective Swallow Measures and Previously Published 'At Risk' Of Airway Violation Reference Threshold Values

Quantitative and objective swallow measures	Reference value
Time to airway closure (Airwaycl)	Above previously published reference mean considered higher risk than average infant with feeding difficulties*
PES opening duration (PESdur)	Below previously published reference mean considered higher risk than average infant with feeding difficulties*
Total pharyngeal transit time (TPT)	Infants 0-9 months: risk of penetration-aspiration twice as likely, when TPT = ≥ 0.5s Children over 9 months: risk of penetration-aspiration increased by 100x when TPT = ≥ 0.2s *,**
Coordination of airway closure with bolus transit (BP1AEcl)	Risk of penetration-aspiration ↑ if BP1 > than 0.1 sec prior to AEcl **
Number of sucks per swallow	>3 sucks per swallow =↑ aspiration risk *,***
Pharyngeal constriction ratio (PCR)	Risk of penetration–aspiration 100 times greater, when PCR = ≥ 0.2 **
Penetration-aspiration scale (PAS) (1-8)	PAS3+ = at risk of penetration-aspiration
Nasopharyngeal redirection (NPR) (present/ absent)	Present = at risk of penetration-aspiration
Pharyngoesophageal redirection (present/ absent)	Present = at risk of penetration-aspiration
Residue (present/ absent)	Present = at risk of penetration- aspiration
Bolus clearance ratio (BCR)	At risk of penetration aspiration if BCR- < 5% **

Notes. *Dharmarathna et al. (2020); ** Dharmarathna et al. (2021); ***Fuller et al. (2022).

For all pertinent swallow measures, all measures obtained from each child's VFSS were compared to previously published 'at risk' threshold values that demonstrate risk of airway violation (Miles et al., 2022). Due to an absence of comparative safety values for airway closure and PES opening duration, data were compared with the mean reported in the previously published mixed aetiology VFSS data (Dharmarathna et al., 2020b). If a child met the 'at risk of airway violation threshold value' for any individual swallow, they were considered at risk.

When analysing measures that were obtained in the three children with repeat (two) VFSS over time, the mean and median values of quantitative VFSS measures and PAS measures were taken from the individual swallows were calculated. This data was then compared across time with reference values from The University of Auckland Swallowing Research Laboratory.

Chapter 5 Results

A total of 20 bottle fed infants with laryngomalacia between 1- and 13 months of age (median = 3 months) met the criteria and were recruited to this retrospective study with VFSSs completed between 2012 and 2022. Table 6 provides a summary of children's demographic and clinical information. The cohort was categorised into three subgroups, namely: 1) laryngomalacia in isolation (laryngomalacia; $n = 6$), 2) laryngomalacia with a concurrent chromosomal or neurological disorder (syndromic; $n = 6$), and 3) laryngomalacia with a concurrent additional anatomic abnormality (anatomic; $n = 8$). Three children received a repeat VFSS which assessed bottle feeding within 14 months of their original VFSS. The medical notes inconsistently reported the severity of laryngomalacia and therefore this information has not been included.

Age, prematurity and gender did not differ across diagnostic subgroups ($p > .05$). Thirteen of 20 children underwent either supraglottoplasty or tracheostomy surgery prior to their initial VFSS. One child received a tracheoesophageal fistula repair prior to VFSS. GERD was documented in half of the children's medical records; the prevalence of GERD was highest in the syndromic subgroup (67%), followed by the anatomic subgroup (50%), although this was not statistically different ($\chi^2 = 1.333$ $p = .513$). Eight children (40%) had a history of recurrent respiratory illness and 19 children (95%) had reported stridor. Long term enteral feeding was frequent with 13 children (65%) requiring either NGT or PEG feeds to support nutritional requirements.

Table 6

Demographic/Clinical Information

	Total cohort (N = 20)	Laryngomalacia (n = 6)	Syndromic (n = 6)	Anatomic (n = 8)	Statistics
Age	Median age: 3 months Range: 1–13 months	1–3 months: 4 4–6 months: 2	1–3 months: 4 6–12 months: 2	1–3 months: 3 4–6 months: 2 6–12 months: 1 12+ months: 2	H = 0.656 p =.72
Gestational age	Preterm: 9 Term: 11	Preterm: 1 Term: 5	Preterm: 4 Term: 2	Preterm: 4 Term: 4	χ^2 = 3.165 p = .205
Gender	Female: 8 (40%) Male: 12 (60%)	Female: 2 (33%) Male: 4 (66%)	Female: 3 (50%) Male: 3 (50%)	Female: 3 (38%) Male: 5 (62%)	χ^2 = 3.83 p = .826
Primary aetiology		Isolated laryngomalacia with no other medical diagnoses: 6	Numerical chromosomal abnormality: 2 Structural chromosomal abnormality: 2 Neurological component (e.g., abnormal MRI): 5	Abnormal anatomical physiology: Oral phase (e.g., base of tongue collapse, micro/retrognathia): 3 Pharyngeal phase (e.g., vocal fold palsy): 1 Oesophageal phase (e.g., tracheoesophageal fistula): 1 Respiratory (e.g., subglottic stenosis, pectus excavatum, tracheomalacia): 3	-
Ethnicity	European: 11 Indian: 2 Māori: 1, Other Asian: 1 Pacific: 5	European: 4 Indian: 1 Māori: 1	European: 2 Indian: 1 Other Asian: 1 Pacific: 2	European: 5 Pacific: 3	χ^2 = 19.111 p = .14
Surgery	Supraglottoplasty: 12 9/12 prior to VFSS1 Tracheostomy: 5 4/5 prior to VFSS1 TOF surgery: 1 None: 2	Supraglottoplasty: 4 3/4 prior toVFSS1 None: 2	Supraglottoplasty: 3 2/3 prior to VFSS1 Tracheostomy: 3 2/3 prior to VFSS1 None: 0	Supraglottoplasty: 5 4/5 prior to VFSS1 Tracheostomy: 2 2/2 prior to VFSS 1 TOF repair: 1 None: 0	Type of surgery- χ^2 = 9.319 p = .156 Surgery pre/post VFSS- χ^2 = 6.161 p = .187

	Total cohort (N = 20)	Laryngomalacia (n = 6)	Syndromic (n = 6)	Anatomic (n = 8)	Statistics
Need for enteral feeding	NGT: 8 PEG: 5 Oral feeding only: 7	NGT: 5 PEG: 0 Oral feeding only: 1	NGT: 1 PEG: 3 Oral feeding only: 2	NGT: 1 PEG: 3 Oral feeding only: 3	$\chi^2 = 8.179$ $p = .085$
Additional aerodigestive medical history	GERD: 10 Documented respiratory concerns: 8 Stridor: 19	GERD: 2 Documented respiratory concerns: 3 Stridor: 5	GERD: 4 Documented respiratory concerns: 2 Stridor: 6	GERD: 4 Documented respiratory concerns: 3 Stridor: 8	GERD – $\chi^2 = 1.333$ $p = .513$ Resp – $\chi^2 = 2.456$ $p = .293$ Stridor – $\chi^2 = 0.382$ $p = .826$

This study's quantitative swallow measures are displayed in Table 7.

Table 7

Descriptive Timing and Displacement Data

Measure		Total cohort (N = 20)	Laryngomalacia (n = 6)	Syndromic (n = 6)	Anatomic (n = 8)	Statistics
Timing measures	PES opening duration (PESdur)	*M* = 0.267 *SD* = 0.127 Range = 0.133–0.53	*M* = 0.363 *SD* = 0.09 Range = 0.287–0.53	*M* = 0.277 *SD* = 0.142 Range = 0.133–0.525	*M* = 0.18 *SD* = 0.09 Range = 0.083–0.375	H = 7.01 *p* = .03
	Time to achieve airway closure (Airwaycl)	*M* = 0.277 *SD* = 0.15 Range = 0.084–0.565	*M* = 0.406 *SD* = 0.157 Range = 0.234–0.565	*M* = 0.271 *SD* = 0.136 Range = 0.129–0.444	*M* = 0.184 *SD* = 0.754 Range = 0.084–0.284	H = 6.810 *p* = .03
	Total pharyngeal transit time (TPT)	*M* = 1.13 *SD* = 0.663 Range = 0.25–2.903	*M* = 1.15 *SD* = 0.326 Range = 0.902–1.575	*M* = 1.38 *SD* = 0.908 Range = 0.860–2.903	*M* = 0.939 *SD* = 0.657 Range = 0.25–2.375	H = 2.724 *p* = .256
	Coordination of airway closure with bolus transit (BP1AEcl)	*M* = 0.113 *SD* = 0.707 Range = 0.00–0.238	*M* = 0.159 *SD* = 0.065 Range = 0.033–0.225	*M* = 0.116 *SD* = 0.089 Range0.008-0.238	*Mean*= 0.077 SD= 0.402 Range: 0.00-0.127	H= 4.107 p= 0.847
	Number of sucks per swallow (SSB ratio)	Median: 2 Range: 1-7	Median: 3 Range: 1-5	Median: 3 Range: 1–7	Median = 2 Range = 1–4	H = 1.717 *p* = .424
Displacement measures	Pharyngeal constriction ratio (PCR)	Median = 0 Range = 0.00– 0.131	Median = 0 Range = 0	Median = 0 Range = 0.00–0.062	Median = 0 Range = 0.00–0.131	H = 0.929 *p* = .629

Note. Bold = $p > .05$.

In Table 8, quantitative measures (timing and displacement measures) are compared to previously published reference threshold values that demonstrate risk of airway violation. Children in the laryngomalacia in isolation subgroup met the at risk of airway violation threshold values more often than children in the other two diagnostics subgroups for all measures except PESdur, Airwaycl, and PCR.

Table 8

Initial VFSS 'At-Risk Of Airway Violation' Timing and Displacement Measures Compared to Previously Published 'At-Risk' Values

Measure + normative value		Total cohort (N = 20)	Laryngomalacia (n = 6)	Syndromic (n = 6)	Anatomical (n = 8)	Comparative previously published mixed aetiology (n = 166)*,**,***
Timing measures	PES opening duration (PESdur) *(below mean = higher risk than average infant with feeding difficulties)**	18/20 = below comparative mean	5/6 = below comparative mean	6/6 = below comparative mean	7/8 = below comparative mean	M = 0.381 SD = 0.214
	Time to airway closure (Airwaycl) *(above mean = higher risk than average infant with feeding difficulties)**	14/20 = above comparative mean	5/6 = above comparative mean	4/6 = above comparative mean	5/8 = above comparative mean	M = 0.110 SD = 0.986
	Total pharyngeal transit time (TPT) *Infants 0-9 months:* *Risk of PA was twice as likely, when TPT = ≥ 0.5s at under 9 months old* *Risk of PA increased by 100x when TPT = ≥ 0.2s at over 9 months old*,***	19/20 = risk of aspiration	6/6 = risk of aspiration	6/6 = risk of aspiration	7/8 = risk of aspiration	M = 0.283 SD = 1.006
	Coordination of airway closure with bolus transit (BP1AEcl)	16/20 = risk of aspiration	6/6 = risk of aspiration	5/6 = risk of aspiration	5/8 = risk of aspiration	M = 0.065 SD = 0.410

Measure + normative value		Total cohort (N = 20)	Laryngomalacia (n = 6)	Syndromic (n = 6)	Anatomical (n = 8)	Comparative previously published mixed aetiology (n = 166)*,**,***
	Risk of PA was ↑if BP1 > than 0.1 sec prior to AEcl **					
	Number of sucks per swallow (SSB ratio) *>3 sucks per swallow =↑aspiration risk* *,***	9/20 = risk of aspiration	3/6 = risk of aspiration	3/6 = risk of aspiration	3/8 = risk of aspiration	
Displacement measures	Pharyngeal constriction ratio (PCR) *Risk of penetration–aspiration* *was 100 times greater, when PCR = ≥ 0.2* **	2/20 = risk of aspiration	0/6 = risk of aspiration	1/6 = risk of aspiration	1/8 = risk of aspiration	M = 0.214 SD = 0.199

Note. *Dharmarathna et al. (2020); ** Dharmarathna et al. (2021); ***Fuller et al. (2022).

Penetration, Aspiration, and Bolus Retention Issues

Measures of airway violation, bolus retention and redirection are displayed in Table 9. Penetration or aspiration was found in 50% of all children across all diagnostic subgroups (PAS3+, *n* = 13). All children in the isolated laryngomalacia subgroup had an episode of penetration or aspiration at some point in their study. This group also had the highest Max PAS score (PAS 6–8 = 100%), when compared to the other diagnostic subgroups but this was not statistically significant. Silent aspiration (PAS 8) captured across the whole study was common with ten out of 13 children who penetrated or aspirated scoring PAS 8. All airway violation events occurred mid swallow.

Table 9

Signs of Swallowing Difficulties Across Cohort

Measures of aspiration, bolus retention and redirection	Total cohort (N = 20 children; n = 147 swallows)	Laryngomalacia (n = 6)	Syndromic (n = 6)	Anatomic (n = 8)	Statistics
Maximum Penetration- aspiration (PAS) across whole study	PAS 1–2: n = 7, 35% PAS 3-8; n =13, 65%	PAS 1–2: n = 0, 0% PAS 3-8: n = 6, 100%	PAS 1–2: n = 2, 33% PAS 3-8: n = 4, 66%	PAS 1–2: n = 5, 63% PAS 3-8: n = 3, 37%	H = 2.161 p = .339
PAS 1–2 = normal range *PAS 3+ = abnormal* *	10/13 = silent aspiration (PAS 8)	5/6 = silent aspiration (PAS 8)	4/4 = silent aspiration (PAS 8)	1/3 = silent aspiration (PAS 8)	
Nasopharyngeal reflux (NPR) (y/n) *No = normal*	Yes: 4 No: 16	Yes: 2 No: 4	Yes: 1 No: 5	Yes: 1 No: 7	χ^2 = 0.99 p =.610
	4/20 = outside normal range	2/6 = outside normal range	1/6 = outside normal range	1/8 = outside normal range	
Residue (y/n) *No = normal*	Yes: 4 (Max BCR range: 10.2–50.4%) No: 16	Yes: 0 No: 6	Yes: 2 (Max BCR range: 10.8–50.4%) No: 4	Yes:2 (Max BCR: 10.2–24.6%) No: 6	χ^2 = 2.92 p = .318
Bolus clearance ratio (BCR)- < 5% *	4/20 = outside normal range	0/6 = outside normal range	2/6 = outside normal range	2/8 = outside normal range	
Pharyngoesophageal redirection (oesophagus – pharynx) (y/n)	Yes: 2 No: 18	Yes: 0 No: 6	Yes: 0 No: 6	Yes: 2 No: 6	χ^2 = 1.019 p = .601
No = normal	2/20 = outside normal range	0/6 = outside normal range	0/6 = outside normal range	2/8 = outside normal range	

Note. *Dharmarathna et al. (2021).

Residue was not significantly correlated with higher PAS scores and residue was less prevalent when compared to penetration or aspiration events [PAS3+ (65%), NPR (20%), EPR (10%), residue (20%)]. The syndromic subgroup had the most residue and largest BCR maximum value (50.8%), although this was not significantly different ($\chi^2 = 2.92$, $p = .318$). Redirection (retrograde movement of bolus from oesophagus back into pharynx) was present in two out of eight children in the anatomic group only.

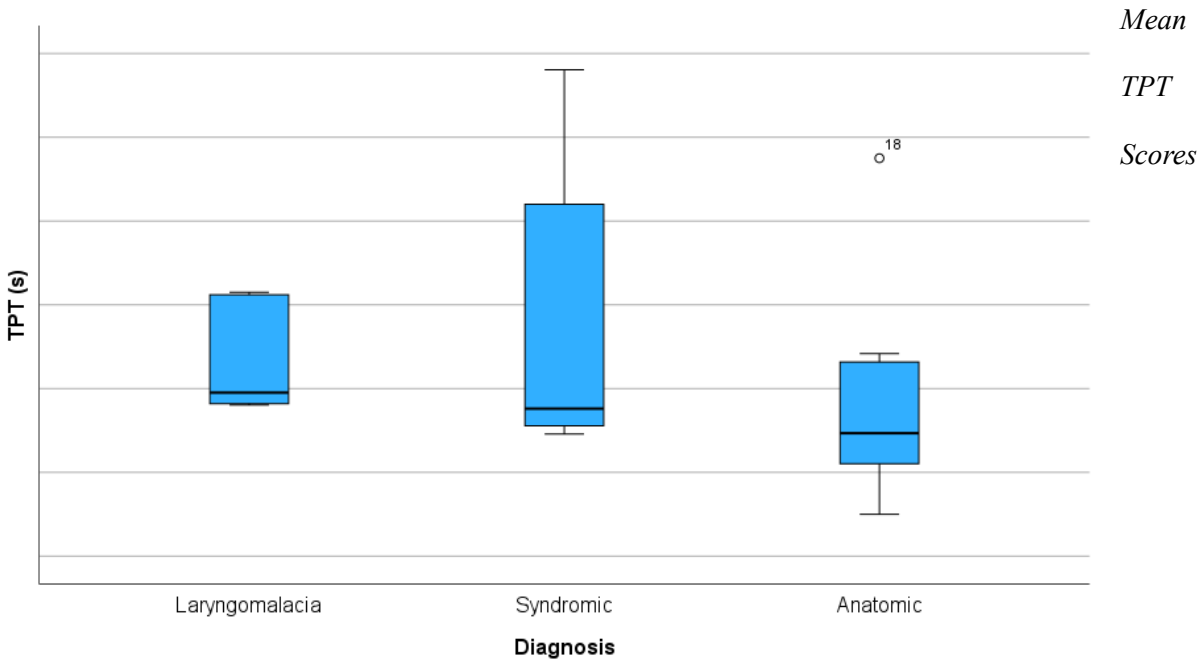
Pharyngeal Constriction Ratio

PCR was not statistically significant across diagnostics subgroups and only two children met the 'at risk of airway' violation threshold values (PCR > 0.2cm^2; Dharmarathna et al., 2021). Child 4 in the syndromic group who was diagnosed with a chromosomal abnormality and global developmental delay had the worst constriction value (0.893) which was followed by Child 15 in the anatomic group who had tracheomalacia (0.239). Elevated PCR (poor constriction) significantly correlated with elevated BCR (increased residue) ($R_s = 1.000$, $p = .01$).

Timing/Coordination Measures

Total Pharyngeal Transit time (TPT) scores were longer in all groups when compared with the 'at risk of airway violation' threshold values (risk of penetration or aspiration twice as likely when TPT = ≥ 0.5s at under 9 months old; Dharmarathna et al., 2021). TPT measures met the at risk threshold values in 19 out of 20 children. The longest TPT maximum value and longest mean TPT was found in the syndromic group; however, this was not significant (mean TPT = 1.38, maximum TPT value = 2.903). TPT was not significantly correlated with any other measure ($p > .05$).

Figure 6



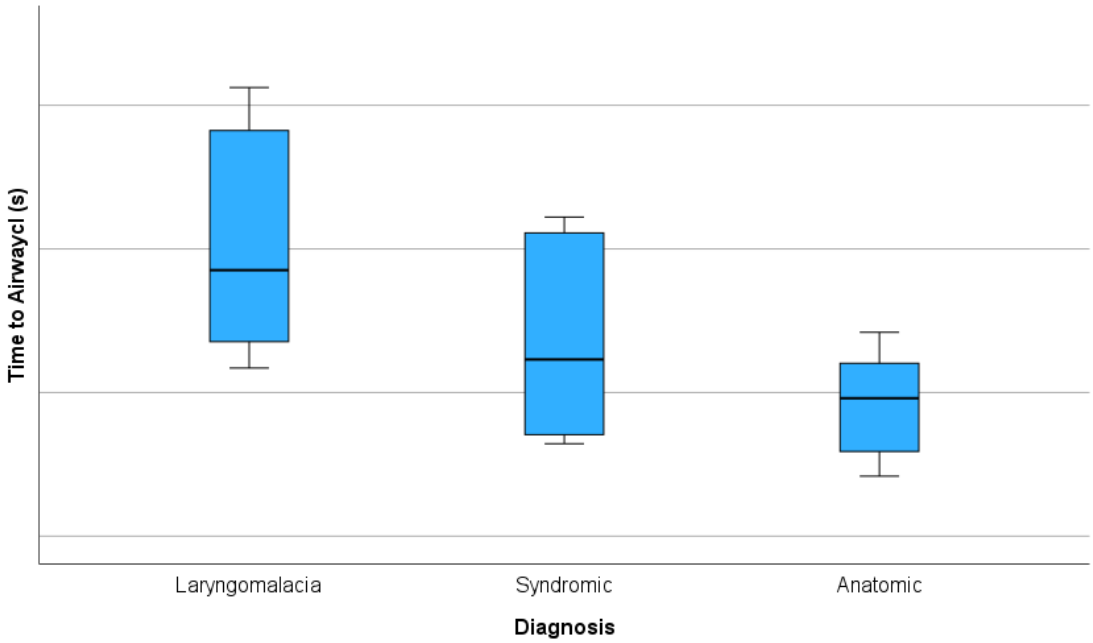
According to the Diagnostic Group

Similarly, 16 out of 20 children had a BP1AEcl score which was longer (longer time with airway open after the bolus reaches the UES, i.e., more at risk of airway violation) than the at-risk threshold values. Elevated BP1AEcl (more at risk) was significantly associated with higher PAS scores ($R_s = 0.643$, $p = .02$).

Time to achieve airway closure (Airwaycl - time taken to reach airway closure from initiation of arytenoid upward movement to full supraglottic closure) was above the previously published mean values in 14 children (Dharmarathna et al. 2020). Airwaycl was statistically associated with elevated PAS scores (Max PAS $R_s = 0.588$, $p = .01$). Time to achieve Airwaycl was highest (i.e., slowest) in the laryngomalacia in isolation subgroup, followed by the syndromic subgroup (see Figure 7). Children in the isolated laryngomalacia group and syndromic subgroup had significantly wider range and longer Airwaycl times when compared to the anatomic subgroup ($H = 6.810$, $p = .03$). Furthermore, children with GERD had statistically significant longer Airwaycl times compared to those without GERD ($H = 4.480$, $p = .034$).

Figure 7

Mean Airwaycl Scores According to the Diagnostic Group



Eighteen out of 20 children had a shorter PES opening duration (PESdur) when compared to previously published mean values (Dharmarathna et al., 2020). Children in the anatomic group's PESdur scores were statistically shorter than the other subgroups (H = 7.01, p = .03).

Nine children in the study had >3 sucks per swallow which is considered to increase the risk of airway violation. The syndromic group showed the greatest range of number of sucks per swallow (range = 1–7). The child with the greatest number of sucks per swallow (n = 7) was Child 13 who had bilateral vocal fold palsy followed by Child 15 who had tracheomalacia (n = 6).

Change in VFSS Metrics Over Time (n = 3)

Despite there being few children with repeat VFSS conducted in the same manner over time, we still felt it worthwhile to follow the trajectory of change. Tables 10, 11 and 12 provide summaries of three children's initial and repeat VFSS that were completed within a period of 14 months. The infants' ages ranged from 3–13 months at initial VFSS and 17–24 months one

month after final VFSS. Two children were in the syndromic group and one child was considered in the anatomic group.

Child 5

Child 5 was diagnosed with severe laryngomalacia, developmental delay, chronic lung disease and had an abnormal MRI. They were 3 months old at the initial VFSS and 17 months old at the final VFSS. Child 5 had a tracheostomy in situ during both studies and was decannulated three years after VFSS 2.

Table 10

Child 5 At-Risk Displacement Measures and Change in Swallowing Outcomes Over Time

Measure	Child 5 (Syndromic group)			
	VFSS 1	Above risk threshold (VFSS 1)	VFSS 2	Above risk threshold (VFSS 2)
TPT	*M* = 2.903	+	*M* = 1.433	+
BP1AEcl	*M* = 0.069	-	*M* = 0.350	+
SSB ratio	Median = 3	+	Median = 1	-
PCR	Median = 0	-	Median = 0	-
Max PAS	8	-	4	+

Note. + = Above risk threshold; - = Not above risk threshold.

In VFSS 1 when the TPT score and SSB ratio were above the at risk of airway violation reference threshold values, Child 5 has an episode of silent aspiration recorded in the study. In VFSS 2, Child 5 also demonstrated several incidences of airway violation (Max PAS = 4). Whilst the TPT score in VFSS 2 was shorter than VFSS 1, the VFSS 2 TPT score continued to be longer when compared to the at risk threshold values, still placing Child 5 at risk of airway violation. The BP1AEcl score was also longer in the VFSS 2 and met the 'at risk' threshold value. The SSB ratio (1:1) did not meet the at risk of airway violation threshold values (SSB 3+) in VFSS 2 and the PCR scores in both studies did not meet the at risk threshold values based on previous publications.

Child 17

Child 17 was diagnosed with severe laryngomalacia, frequent respiratory illnesses and Beckwith–Wiedemann syndrome. They were 1 year old during the initial VFSS and 2 years 1 month at the second VFSS. Child 17 had a tracheostomy in situ during both studies and was still not decannulated at the point of medical record extraction (age 6 years old).

Table 11

Child 17 At-Risk Displacement Measures and Change in Swallowing Outcomes Over Time

Measure	Child 17 (Syndromic group)			
	VFSS 1	Above risk threshold (VFSS 1)	VFSS 2	Above risk threshold (VFSS 2)
TPT	*M*= 0.729	+	*M* = 1.292	+
BP1AEcl	*M*= 0.212	+	*M* = 0.334	+
SSB ratio	Median = 1	-	Median = 2	-
PCR	Median = 0	-	Median = 0.097	-
Max PAS	8	+	2	-

Note. + = Above risk threshold; - = Not above risk threshold.

In VFSS 1 when the TPT score and BP1AEcl were above the at risk of airway violation reference threshold values, Child 17 had several incidences of airway violation (Max PAS = 8). However, in VFSS 2, Child 17 did not receive a PAS score that met with this study's airway violation values (PAS 3+), yet the TPT and BP1AEcl values still met the 'at risk' threshold values. The pharyngeal transit times (TPT) in VFSS 2 were longer than at VFSS 1 as was the BP1AEcl score. The PCR increased (worsened) at VFSS 2 although this was still below the at risk threshold value of ≥ 0.2.

Child 10

Child 10 was in the 'other anatomical' group; they had moderate laryngomalacia, left vocal fold palsy, base of tongue collapse, bronchiectasis and underwent supraglottoplasty surgery prior to VFSS 1. Child 10 continued to have recurrent respiratory illnesses until they received a third swallow study the following year which was not included as the assessment did not include bottle feeding. This child was subsequently lost to follow-up by the tertiary centre.

Table 12

Child 10 At-Risk Displacement Measures and Change in Swallowing Outcomes Over Time

Measure	Child 10 (Anatomic group)			
	VFSS 1	Above risk threshold (VFSS 1)	VFSS 2	Above risk threshold (VFSS 2)
TPT	*M*= 0.75	+	*M* = 1.397	+
BP1AEcl	*M*= 0.083	-	*M*= 0.084	-
SSB ratio	Median = 1	-	Median = 2	-
PCR	Median = 0	-	Median = 0	-
Max PAS	8	+	1	-

Note. + = Above risk threshold; - = Not above risk threshold.

In VFSS 1 when the TPT scores were above the at risk of airway violation reference threshold values, Child 10 had several incidences of airway violation (Max PAS = 8). However, in VFSS 2, Child 10 did not receive a PAS score that met with this study's airway violation values (PAS 3+). BP1AEcl did not meet the 'at risk' threshold value in either study. The pharyngeal transit times (TPT) in VFSS 2 were longer than at VFSS 1. The PCR and SSB did not meet the 'at risk' threshold values for either study.

Interestingly, during Child 10's third VFSS, aspiration was noted (Max PAS = 7). The BP1AEcl score had increased to 0.094, although this is still below the at risk of airway violation threshold value (risk of penetration or aspiration increases when BP1 > than 0.1 sec prior to AEcl; Dharmarathna et al., 2021).

Chapter 6 Discussion

This study examined swallowing performance of children with laryngomalacia using fluoroscopic quantitative measures. Children with laryngomalacia demonstrated swallowing timing impairments and airway risk even after surgery. Quantitative swallow measures allow the clinician to explore the underlying biomechanical cause of aspiration and support diagnostic decision-making regarding concurrent comorbidities as well as guide targeted interventions.

Demographics

In this tertiary centre, there was a low referral rate to speech-language pathology (26%) and an even lower rate of referral for an VFSS (3%). This may suggest clinicians are either reluctant to refer for a radiological swallow examination when most cases are known to be self-limiting or do not feel that there are swallowing concerns that warrant further investigation. There is a paucity of international data regarding referral rates to Speech and language pathology or instrumental assessment in children with laryngomalacia yet given the high rates of silent aspiration in those identified with swallowing problems apparent in this study and other published studies, a standardised instrumental swallowing assessment protocol may be warranted both pre- and post-surgery for those children identified as struggling or at high risk. Children receiving an instrumental assessment in this study did have overt swallowing concerns after SLT assessment, but they may not be representative of the wider laryngomalacia population within a hospital or outpatients setting. The true prevalence of children with laryngomalacia and feeding difficulties is difficult to ascertain. One factor contributing to the unknown prevalence is that most cases of laryngomalacia are mild and self-resolving and so most children are largely managed by their paediatrician and do not receive SLT review or referral for VFSS (Landry & Thompson, 2017).

This study population from Auckland, New Zealand, demonstrates characteristics similar to previously reported cohorts of children with laryngomalacia. There is a general male predominance found in infants with laryngomalacia (Cooper et al., 2014), and this study found that 60% of infants referred for VFSS were male. Prematurity did not feature as a significant risk factor for laryngomalacia in this study, which aligned with the inconclusive published data regarding the association between laryngomalacia and gestational age (Edmondson et al., 2011). The ethnic diversity of our cohort is reflective of the Auckland, New Zealand population, with the largest group identifying as European, followed by Pacific and Asian (Gilbertson, 2013). Previously published data suggest that there is a higher incidence of laryngomalacia in Hispanic and Black infants (Edmondson et al., 2011), and whilst this study did not contain these specific ethnic groups, our study did not find a similar prevalence of Asian and ethnic minority infants who received a VFSS (45%) when compared to Black, Asian and ethnic minority (BAME) ethnicity data from the Auckland population census (46%) (Gilbertson, 2013).

Our study also had a similar incidence of children with comorbidities (70%) when compared to Irace et al. (2019) (83.8%). Common comorbidities found alongside laryngomalacia include neurologic disease, cardiopulmonary disease, congenital anomalies, neonatal abstinence syndrome (NAS) and other syndromes (Abraham et al., 2022; Thompson, 2007). All of these conditions except NAS were found in our study cohort. Furthermore, 25% of this study's cohort presents with a neurological component, congruent with the previously reported 20% of laryngomalacia cases presenting with neurological conditions, with resolution of symptoms observed following resolution of the neurological condition (Abraham et al., 2022; Thompson, 2007). This study found only 35% of the total cohort required a follow-up instrumental assessment, suggesting there may be some resolution of swallow impairment.

Swallowing Function and Supraglottoplasty

The incidence of surgery and specifically supraglottoplasty within our study was higher than previously published data. Our data showed 85% of children who received a VFSS had surgery, with 60% undergoing supraglottoplasty compared to published data suggesting that 10 to 25% of children who initially present with moderate symptoms require surgery (Bedwell & Zalzal, 2016; Sidell & Messner, 2021). The increased surgical rates further highlight the severity of laryngomalacia in this specific cohort. In our children, tracheostomies were only placed in those with additional syndromic or anatomic abnormalities. Considering the significant comorbidities present in these children we infer that tracheostomy is chosen over supraglottoplasty in these groups, in line with published data suggesting supraglottoplasty may be less effective in patients with multiple comorbidities (Carter et al., 2016; Simons et al., 2016). The evidence surrounding swallow improvement following supraglottoplasty is conflicting, but it is acknowledged that swallowing in children with multiple comorbidities may not improve post-operatively (Jadcherla, 2017; Moroco & Aaronson, 2022; Schroeder et al., 2008). Seventy-six per cent of children had surgery prior to the initial VFSS and ten out of 13 children who had an incidence of airway violation captured at their initial VFSS had undergone supraglottoplasty prior to their swallow study. This study does not have pre- and post-operative VFSS data available for comparison of each child; however, the results available suggest that supraglottoplasty does not completely resolve impaired swallowing biomechanics, particularly when comorbidities or significant swallowing difficulties are present prior to surgery. Surgery does not address sensory issues which may also contribute to worse swallowing metrics and may go unidentified without instrumental assessment (Klinginsmith et al., 2019).

It is possible that surgery further alters sensory mechanisms at the laryngeal inlet, reducing the ability of the airway to respond to bolus material and achieve full aryepiglottic closure in a timely manner. This is evidenced by 1) high rates of silent aspiration, 2) all airway

violation events occurring mid swallow and 3) elevated BP1AEcl findings which placed 16 children in the 'at risk of airway violation' threshold values. Gan et al. (2021) also found evidence to support the altered sensory mechanisms theory in children with laryngomalacia, with 70.5% of children who underwent aryepiglottoplasty showing signs of inflammation of the laryngeal mucosa as well as immature cartilage which may contribute to prolapse of the supraglottic structures during the swallow. The specimens in the same study were taken during surgery prior to symptom resolution and therefore these differences may be short lived.

In our study, all VFSS completed postoperatively were at least 48 hours post-surgery and typically more than one month postoperatively. This is longer than the time to assessment in Rastatter et al. (2010) who completed at least one clinical feeding examination within 48 hours post-surgery. They report a similar rate of postoperative aspiration, 21 out of 39 children (53%), although it is important to note that their study did not report aspiration using PAS scales. Our findings suggests that postoperative aspiration may continue for a lengthier period of time; however, our study also included children with tracheostomies and so direct comparison is not possible.

Previously published data suggests that laryngomalacia is worsened by reflux disease and that children with neurodisabilities have higher incidences of GERD (Trinick et al., 2012). GERD was common in this cohort, yet pharyngoesophageal redirection (reflux) was only captured in two children. VFSS is not a study to diagnose reflux and capturing reflux events on VFSS is often opportunistic. GERD reported in the medical records was not statistically correlated with diagnostic subgroups, which may be due to sample size, and further investigation is warranted. Nevertheless, our findings did not support the hypothesis that bolus redirection is prevalent in children with laryngomalacia, with or without a concurrent chromosomal or neurological component.

Whilst GERD may not be prevalent in a specific subgroup it is associated with 'at risk of airway violation' VFSS timing measures. Children with reported GERD had statistically significantly longer time to achieve Airwaycl ($p = .034$), suggesting that reflux does co-exist with altered swallow biomechanics in this population. It is known that GERD can reduce pharyngeal and tracheal sensitivity in the paediatric population (Link, 2000) and previous authors have proposed that the correlation between time to achieve Airwaycl and GERD may be due to the resistant airway worsening reflux (Hysinger, 2018; Klinginsmith et al., 2019). However, it is also possible that effortful breathing in these infants results in greater reflux through negative pressure generation in the thoracic cavity drawing gastric content in a retrograde manner. Perhaps the reported neurogenic element associated with GERD and resistant airways seen in laryngomalacia significantly impact swallow biomechanics in a manner that this study is unable to fully report for two reasons. We did not have available pre- and post-operative VFSS data and the quantitative VFSS measures do not provide a clear picture of the respiratory aspect in terms of full visualisation of supraglottic structures to fully discuss the association between swallow and respiratory biomechanics. Evaluating quantitative VFSS measures pre- and post-surgery in conjunction with a FEES assessment to observe laryngeal motion, could be a key factor in understanding this relationship

Aspiration

Aspiration and silent aspiration in children with laryngomalacia are commonly reported symptoms (Irace et al., 2019; Richter et al., 2009; Simons et al., 2016). Our findings are similar to Simons et al. (2016), with high rates of aspiration in children with laryngomalacia in isolation, children with a concurrent chromosomal/neurological component or children with a concurrent additional anatomic abnormality. Our study is the first to examine aspiration and residue in relation to specific quantitative VFSS measures in children with laryngomalacia and

we believe that these measures allow us to identify biomechanical changes that cause these events.

It has been hypothesised that laryngomalacia has a sensorimotor cause or exhibits a sensory deficit, and that this should be considered in relation to silent aspiration in this cohort. Children with neurologic impairments often silently aspirate (Weir et al., 2011). Silent aspiration was present in this study (n = 10, 50%) and the greatest PAS scores were found in the laryngomalacia in isolation and anatomic subgroups. The true prevalence of silent aspiration in children with laryngomalacia is unknown due to the lack of routine use of instrumental swallow assessments (Chadha, 2019). Clinical feeding evaluations may not give a complete picture of airway risk, as they will miss silent aspiration events. VFSS is a short study and is a 'snapshot' of the child at one moment; therefore, it cannot capture all possible swallowing patterns. Airway violation events may not be detected on single VFSS assessments because these are limited in duration to manage radiation exposure risks, and because aspiration is a sporadic occurrence (Leonard, 2019). However, additional data captured on VFSS can point to risks of aspiration events when analysed against known quantitative measures. If VFSS is performed and analysed in a systematic fashion, describing physiology, indications of airway risk can be obtained that may still prove valuable when planning management strategies (Leonard, 2019). VFSS should not be regarded as a 'detection' study, looking for aspiration or reflux, but should be seen as an opportunity to evaluate deglutition carefully and produce recommendations based upon current documented function. It is also important to note that the VFSS radiology environment is unable to completely mimic regular infant feeding settings (i.e., positioning, place, feeder, feed time, full volume, and timing), potentially influencing the identification of events and further highlighting the need for calculating quantitative measures.

Pharyngeal Constriction Ratio and Residue

PCR provides information about the overall integrity of pharyngeal constriction, that is, the pharynx's ability to constrict behind the bolus to clear it through the upper oesophageal sphincter. PCR was not statistically different across diagnostics subgroups, disproving our hypothesis that displacement measures and bolus retention may be more prevalent in children with concurrent chromosomal or neurological components. Only two children met the 'at risk of airway violation' threshold values and these children were found in the syndromic and anatomic groups. Dharmarathna et al. (2021) found that PCR had the strongest predictive relationship for airway violation when compared to all other quantitative measures in a mixed aetiology cohort; yet this was not the case in our laryngomalacia cohort. PCR and BCR measures become elevated when weakness is present, and the lack of abnormal measures coupled with lack of correlation with PAS scores, suggests that in this cohort, pharyngeal weakness was not the driving factor for airway intrusion. Further investigation in a larger group of laryngomalacia children is required to ascertain whether PCR and BCR are linked to or can predict swallowing risk in children with laryngomalacia.

Timing Measures

In contrast airway timing measures were associated with risk behaviours. The author hypothesised that there would be a correlation between quantitative timing measures on VFSS and penetration or aspiration (PAS score). Timing measures (time to achieve Airwaycl and BP1AEcl) significantly correlated with airway violation events ($p > .05$). This may reflect sensorimotor integration impairment or a speed issue (in closing the airway). Delayed airway closure was found and did correlate with intrusion, but other quantitative timing measures were not linked to airway violation in non-parametric testing. Airwaycl and PESdur differed significantly when comparing diagnostic subgroups which was less expected and refuted our hypothesis, 'that quantitative airway timing measures will be similar in all children with

laryngomalacia'. Time to achieve airway closure is a timing measure from the point of onset to completion of arytenoid-epiglottis approximation, indicating supraglottic airway closure (Leonard, 2019). There are no published at risk of airway violation threshold values for this measure in children. However, it is thought that a shorter time to achieve Airwaycl would decrease the risk of airway violation as the bolus is less able to enter the airway. Therefore, this study assessed the relative risk of airway violation by comparing these Airwaycl measures with previously published means (Dharmarathna et al., 2020).

Children in the isolated laryngomalacia and syndromic subgroups had significantly wider range and longer Airwaycl times when compared to the anatomic subgroup. The longer time to achieve Airwaycl coupled with intra-deglutitive airway violation is posited to be a result of the physical collapse of tissues obstructing the ability to fully close the airway tightly. This may be a limitation of using VFSS as an instrumental assessment because the increased closure times may falsely indicate that the supraglottic airway closure had occurred through normal non-volitional medullary swallow control, whereas it is a factor of mechanical collapse of supraglottic structures in a haphazard way.

Sixteen out of 20 children had a BP1AEcl score which was longer than the 'at risk of airway violation' threshold values and BP1AEcl was statistically associated with airway violation events ($R_s = 0.688$, $p = .01$). This is unsurprising because children are at higher risk for airway intrusion when the head of the bolus arrives at the PES prior to airway closure (BP1AEcl) due to increased chance of misdirection (Dharmarathna et al., 2020). In typical swallows, the epiglottis approximates the arytenoid cartilages (AEcl) when the bolus arrives in the oesophageal sphincter. Aspiration risk increases if the bolus arrives at the PES less than 0.1 seconds prior to airway closure (Dharmarathna et al., 2021). The prolonged BP1AEcl timings may be due to prolapsed supraglottic structures obstructing true airway closure, which would be

in keeping with our observation that all airway violation events occurred mid-swallow (intradeglutitively).

Alternatively, differences between groups may relate to impacts of other comorbidities or be affected by lower power given the size of each subgroup. Nevertheless, our overall data suggests that aberrant swallow timing measures may contribute to the prevalence of PFD in laryngomalacia.

Total Pharyngeal Transit Time (TPT)

Mean TPT scores for 19 children (95%) met the 'at risk of airway violation' threshold values yet penetration or aspiration (PAS 3+) was only observed in 65% of children. The risks associated with prolonged bolus transit times are well documented in both adult and paediatric population studies (Dharmarathna et al., 2021; Leonard, 2019). Lingering bolus material in the pharynx poses a greater challenge to airway protective mechanisms (i.e., aryepiglottic closure, hyoid elevation etc.). Irrespective of airway violation events, in an adult population study, prolonged TPT scores were associated with aspiration pneumonia even when aspiration was not documented during the study (Johnson & McKenzie, 1993). Therefore, it is hypothesised that increased TPT measures infer a risk in this paediatric population over time, irrespective of identified airway violation events on individual VFSS.

PES Opening Duration (PESdur)

Similarly to the TPT values, 18 out of 20 children had a shorter PES opening duration (PESdur) when compared to previously published mean values (Dharmarathna et al., 2020) increasing the risk of residue accumulating that could then misdirect into the airway. However, we did not find elevated residue scores and despite children in the anatomic group showing PESdur measures that were statistically shorter than the other diagnostic subgroups ($H = 7.01$, $p = .03$) there was no correlation with airway violation events or GERD. Early closure of the PES may be due to weakness in holding the larynx in an elevated position, or a sensory issue with

incorrect detection of bolus size, and thus inadequate modulation occurring to enable full bolus passage. Therefore, this metric could support *either* a weakness paradigm or a sensorimotor impairment underlying laryngomalacia.

Suck Swallow Breath Ratio (SSB)

Nutritive sucking is a neurobehavioral sensorimotor activity (Ross & Fuhrmann, 2015). Nine children in the study had more than three sucks per swallow which is considered to increase the risk of airway violation. Children in the syndromic group showed the greatest range of number of sucks per swallow (range = 1–7) which is to be expected due to underlying neurologic comorbidities. Whilst not statistically correlated with airway violation events, the large number of children with more than three sucks per swallow may align with the neurologic dysfunction theory in laryngomalacia and suggests that children with laryngomalacia may have mild laryngeal neurological impairment. This theory is also supported by a rising incidence of neurologic impairments reported as the severity of laryngomalacia increases (Klinginsmith et al., 2019). Alternatively, a sensory impairment proposed by Gan et al. (2021) may impact the detection of volume collecting in the pharynx leading to a risk of aspiration; this still supports a neurologic dysfunction theory whilst also including the specific sensory dysfunction seen in children with laryngomalacia.

Change in Swallowing Outcomes Over Time

Laryngomalacia often spontaneously resolves by 12–18 months old (Olney et al., 1999) although this was not the case for all children in this study. Of 877 children coded with laryngomalacia, only three children were identified as having a repeat VFSS using the same feeding method to allow for comparison of studies over time. This limits any conclusions that can be drawn about the trajectory of swallowing change in laryngomalacia children. This follow-up cohort should also be viewed with caution because ages ranged from 3 months – 25 months which creates variability in measures due to naturally changing and developing anatomy

(Morris & Klein, 2000) and developmental changes including positioning changes during feeding, recommended volumes and age-appropriate equipment for fluid intake. Timing measures continue to predict airway violation events in the three repeat VFSS exams available for analysis however presence of tracheostomy was noted in two children. Diminished laryngeal elevation associated with tracheostomies (Abraham & Wolf, 2000) may explain longer BP1AEcl timing as it is harder to elevate the larynx to meet the epiglottis with the tube in place. TPT scores increased over time for Child 10 and Child 17 which may be expected as the relative size of the pharynx increases with age, the larynx descends, elongating and expanding the pharynx (Logemann, 1998). Furthermore, the presence of a tracheostomy and prolonged TPT scores may be due to delayed laryngeal elevation seen in patients with tracheostomies (Abraham & Wolf, 2000). Regardless of captured aspiration events on VFSS, prolonged TPT scores are known to increase the risk of aspiration in adults. A larger prospective, longitudinal study with repeated quantitative VFSS measures is required to provide further information regarding swallowing trajectory.

Comorbidities and Swallowing Outcomes

Frequent comorbidities are found in infants with laryngomalacia, influencing swallowing outcomes and making it difficult to establish direct risk of laryngomalacia alone. There were an increased number of gastrostomy insertions in the syndromic and anatomic subgroups (43%) when compared with the 'laryngomalacia in isolation' group who had none. Repeated VFSS for these children may have been undertaken to evaluate whether shared feeding (oral and tube fed) could be instituted. These decisions would need to be made on a case-by-case basis.

Other published work suggests that laryngomalacia in infancy affects laryngeal structures into adolescence (Hilland et al., 2016). Our study data presents quantitative swallow measures (TPT, BP1AEcl) at two time points, that continue to reach risk thresholds for increased 'risk of airway violation' over time, suggesting in some children that there is an

ongoing risk. Clinicians should consider repeating instrumental swallow assessments when clinically indicated and include calculating quantitative swallow timing measures for children with a history of laryngomalacia and continued aerodigestive problems.

Limitations

We acknowledge several limitations of this work.

Sample Size

The cohort size in this study was small (n = 20). This may be attributed to the single centre site used for recruitment, reluctance to send young children to the video suite and expose them to radiation, or to spontaneous resolution of laryngomalacia limiting the number of referrals for VFSS despite the prevalence of feeding difficulties in infancy (Irace et al., 2019; Simons et al., 2016). The small sample size did not provide adequate power to identify statistically significant changes in objective measures or in swallow patterns over time. The follow-up cohort (n = 3) was very small and did not allow for statistical analysis, and results from this data should be interpreted with caution. Furthermore, in our cohort, a wide range of additional disorders were coded alongside the diagnosis of laryngomalacia which may also have impacted swallowing. This further limits the ability to identify specific swallowing measures that are unique to laryngomalacia or contribute to PFD.

Selection Bias

All children recruited for this study were reported to present concerns with feeding. In this study, the rate of penetration was elevated, which may reflect the selection of children with worse swallowing profiles. Because of this inherent selection bias, the swallow biomechanics reported herein may not apply to a wider laryngomalacia population. Furthermore, due to retrospective tertiary centre recruitment, a large proportion of the children in this study have comorbidities, making it difficult to extrapolate the data to a wider cohort who may differ in the presence of other disease or disorders.

Lack of pre- and post-operative measures prevented evaluation of surgical effects on swallowing. Prolapse of supraglottic tissues affects airway function in these children but somewhat ironically, the prolapsed tissue prevents normal airway closure from occurring, so does not seem to offer normal protection from bolus airway intrusion.

Videofluoroscopic Swallow Study Radiation, Video Quality, and Retrospective Analysis

Due to the ionising radiation delivered during VFSS and considerations required when exposing children to radiation, recordings are limited in time and frame rate compared to adult protocols (Miles et al., 2022). Therefore, some instances of swallow impairments, specifically when observing bottle-fed infants, may not have been recorded due to the limited length of video loops and quality (McGrattan et al., 2020).

The retrospective design of this study means bolus size, time of feed (i.e., mid feed sucking) or teat type (brand and flow rate) could not be controlled. Labelling of VFSS loops taken from the University of Auckland Videofluoroscopy Database for Children was also unclear regarding fluid consistencies and teat type. Therefore, there may be variability in bolus viscosity affecting the analysis. Calibration rings were present in only 2 out of 20 of the total cohort, making calculation of displacement measures such as PESmax impossible. Whilst this limits the analysis, the BCR and residue data suggest that displacement measures were not as significantly affected within this cohort as timing measures. These factors should be considered when recording new VFSSs for future analysis.

Normative Data

Due to the risks of VFSSs in children, no normative paediatric reference values are currently available in the literature for any biomechanical swallow measures. Therefore, a comparison was made to previously published mixed aetiology data from the Auckland University Swallow Laboratory. Published cut-off reference values were used to compare the current study's biomechanical swallow measurements and airway risks. There are currently no

reference values in the literature for airway closure (Airwaycl) or PES opening duration (PESdur); therefore, mean values from previously published paediatric data were used. This limits the analysis and conclusions that can be drawn. Dharmarathna and colleagues (2020a, 2020b, 2021) have compiled biomechanical swallow data for 553 children referred for a VFSS due to PFD concerns, creating representative paediatric data to predict aspiration risk. Whilst this is not a normative cohort, it provides researchers with the ability to predict aspiration events using data from a mixed aetiology cohort (neurological, chromosomal, anatomical, respiratory, cardiac, gastrointestinal, multiple/unknown aetiologies) (Dharmarathna et al., 2021). Thus, there are limitations when comparing laryngomalacia swallowing measures to a mixed aetiology PFD cohort.

Future Directions

Future research should evaluate a larger cohort of children with laryngomalacia using quantitative swallowing measures in a prospective longitudinal study to assess whether swallowing change follows the purported natural resolution over time that laryngeal changes are said to make. Studies should consider the effects of environmental factors such as positioning, bolus volume, and temporal changes. Particular focus should be placed on VFSS data in children with mild laryngomalacia as well as pre- and post-operatively and at specific time periods (e.g., following the introduction of solids, cup drinking).

Conclusion

The present study used quantitative videofluoroscopic swallowing measures to examine the swallow biomechanics of bottle fed children with laryngomalacia with and without coexisting comorbidities using published paediatric fluoroscopic measures for comparison. Only 3% of children with laryngomalacia were referred for a VFSS over 10 years at a tertiary centre. The majority of children referred had multiple comorbidities alongside laryngomalacia and required instrumental assessment which identified significant swallowing impairments.

This preliminary study is the first to examine aspiration and residue in children with laryngomalacia by using specific quantitative VFSS measures. Airway violation, increased time to airway closure and delayed airway closure in relation to bolus position were common in those with laryngomalacia in isolation as well as those with other comorbidities even after supraglottoplasty. Impaired pharyngeal constriction and pharyngeal residue were uncommon suggesting that impairments affected sensory and timing issues to a greater extent. Longer time to achieve airway closure coupled with intra-deglutitive airway violation is posited to be a result of the physical collapse of tissues obstructing the ability to fully close the airway tightly which may be a limitation of using VFSS as an instrumental assessment alone. It is hypothesised that supraglottic airway closure may not fully occur due to mechanical collapse of supraglottic structures in a haphazard way.

VFSS may not be required for all children with laryngomalacia, but swallow dysfunction should be considered in those with respiratory or feeding concerns or other comorbidities. Penetration or aspiration captured during studies should not be the only measure considered when making clinical decisions regarding management. For children who undergo instrumental assessment, using quantitative measures allows the clinician to predict risk, as well as providing an overall holistic picture of the child's swallowing function.

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Appendix A

Quantitative Measures*

Objective measure	Definition	Reference values
Timing(s)/Coordination		
Total pharyngeal transit time (TPT)	Represents the total time of the bolus passage through the pharynx, from when the bolus head passes the posterior nasal spine (BP1) to the time at which the bolus tail completely clears the PES (BP2). *Total pharyngeal transit time = BP2-BP1* *https://youtu.be/p9Ayt6DfQoo*	*Children: Risk of penetration– aspiration was 100 times greater when TPT = ≥ 2s (Dharmarathna et al., 2021).* *Infants: Risk of penetration– aspiration was twice as likely, when TPT = ≥ 0.5s (Dharmarathna et al., 2020).*
Time to airway closure	Time taken to total arytenoid-epiglottis approximation to close supraglottic airway. *Airway start (AEs)-airway close (Acl)* *https://youtu.be/IS1_C4k473Q*	*BCR significantly associated with Time to airway closure (Dharmarathna et al., 2021).*
PES opening duration (PESdur)	The duration of PES opening from the first frame in which it opens (Pop), to when it closes behind the bolus tail (Pcl). *PES opening time = Pcl-Pop* *https://youtu.be/Uh0BKXNF-us*	*Shorten PESdur = τ aspiration risk (Dharmarathna et al., 2020).* *BCR significantly associated with PESdur (Dharmarathna et al., 2021).*
Coordination of airway closure with bolus transit	Airway closure time (Aec) in relation to bolus reaching PES (BP1). *Coordination of airway closure with bolus transit = BP1-Acl*	*PAS scores are higher, when the bolus arrives at the PES prior to airway closure (Dharmarathna et al., 2020).*
Number of sucks-per-swallow	*Downward motion of mandible-to-mandible returning to neutral position was counted as one suck. Total number of sucks per swallow was counted.*	*>3 sucks per swallow = τ aspiration risk (Dharmarathna et al., 2020).*
Displacement measures (cm)		
Pharyngeal constriction ratio (PCR)	The ratio of pharyngeal area at maximum constriction to the area of the pharynx at rest. *PCR = Maximum pharyngeal area divided by maximum pharyngeal constriction.* *https://youtu.be/f17HH7xyJHM*	*Risk of penetration–aspiration was 100 times greater when PCR = ≥ 0.2 (Dharmarathna et al., 2021).* *PCR of 0.2 or higher (worse) was more likely to demonstrate NPR (Dharmarathna et al., 2020).* *τPCR= τEPR risk (Dharmarathna et al., 2020).*
PES max opening (PESmax) *needs calibration ring	The width of the pharyngoesophageal segment was measured at the point of maximum opening during the swallow.	*τPESmax = -τNPR risk (Dharmarathna et al., 2020).*

Objective measure	Definition	Reference values
Bolus clearance ratio	*https://youtu.be/Soh0UlhOa-c*	*BCR significantly associated with PCR, TPT, time to airway closure and PESdur (Dharmarathna et al., 2021).* *Risk of penetration–aspiration was 100 times greater when BCR = ≥ 0.1 (Dharmarathna et al., 2021).* *There was a 20-fold increased risk of penetration–aspiration when BCR was increased by one point (Dharmarathna et al., 2021).* *τ BCR = τaspiration risk (Dharmarathna et al., 2020).* *BCR > 0.3 = τ EPR risk (Dharmarathna et al., 2020).*
Subjective measures		
Penetration- aspiration scale	1–8	> 3 indicates airway violation
Frequency of aspiration	Number of times a bolus passed below the vocal folds in a 20 second loop	τ frequency = τ aspiration risk
Naso pharyngeal regurgitation (NPR)	Presence or absence of NPR marked as (+) or (-)	+ = τ aspiration risk
Pharyngo-esophageal regurgitation (PER)	Presence or absence of PER, marked as (+) or (-)	+ = τ aspiration risk

Notes. s = seconds; cm = centimetres

In children with several measures elevated, the risk of penetration–aspiration climbed steeply, with a 100 times greater risk in those with combined presence of elevated bolus clearance ratio, prolonged pharyngeal transit time, poor pharyngeal constriction, and delay in maximum hyoid elevation (Dharmarathna et al., 2021).

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