Laryngeal ultrasound for evaluation of paediatric unilateral vocal fold immobility – a retrospective cohort study in Aotearoa New Zealand and a survey of clinicians around the world

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Abstract

Objective: Vocal fold immobility (VFI) is a cause of significant morbidity and mortality in the paediatric population. Laryngoscopy is the current first-line investigation for patients with suspected VFI. Laryngeal ultrasound (LUS) has recently emerged as an alternative method of identifying VFI. Compared to laryngoscopy, LUS is less invasive, does not require anaesthesia, and can be performed by non-otolaryngologists. The primary objective of this study is to evaluate LUS as a diagnostic method for the identification of unilateral VFI in a cohort of paediatric patients in Aotearoa New Zealand (NZ). The secondary objective is to estimate the frequency of use of LUS in the paediatric population around the world.

Methods: A retrospective, single-centre cohort study was performed on all paediatric patients who had undergone laryngoscopy and LUS at Starship Children’s Health between 2020 to 2023 in Auckland, NZ. An eight-question survey was also developed and distributed to better understand clinicians’ use of LUS to diagnose paediatric VFI globally.

Results: Twenty-three paediatric patients met the inclusion criteria. LUS demonstrated high sensitivity (100%), specificity (63%), positive predictive value (83%), and negative predictive value (100%) for detecting unilateral VFI within our patient population. Out of the eighty-seven respondents to the survey, nearly half utilise LUS in their clinical practice in the paediatric population. The main barrier for those who do not is lack of expertise, equipment, and training.

Conclusions: These findings support the use of LUS as a safe and accurate diagnostic tool for the detection of unilateral VFI. Further studies in non-surgical populations and standardised guidelines for LUS technique and reporting are required.

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Key Points

Laryngeal ultrasound (LUS) has recently emerged as a non-invasive and reproducible additional method of diagnosing paediatric vocal fold immobility (VFI) relative to laryngoscopy.

This is the first study to investigate the diagnostic accuracy of LUS at identifying unilateral VFI in paediatric patients in Aotearoa New Zealand.

LUS demonstrated high sensitivity and specificity for the detection of unilateral VFI within our cohort.

The study survey demonstrated an increasing clinical interest in the use of LUS for the detection of VFI. Lack of expertise and equipment were considered the main barriers to achieving this.

Further research into the scope, training requirements, and interpretation of LUS is required to validate its use as a point-of-care diagnostic test for paediatric VFI.

Keywords

Vocal fold movement impairment; vocal cord paralysis; flexible laryngoscopy; larynx; laryngeal ultrasound; sonography; paediatric otolaryngology; paediatric airway.

Introduction

Vocal fold immobility (VFI) is the second most common cause of stridor in paediatric patients (1-3). Significant morbidity and mortality are associated with VFI, with increased risk of aspiration and airway obstruction. The gold standard for diagnosing VFI is laryngoscopy. The latter, however, is not without risk and is an aerosol-generating procedure. Respiratory distress, hypoxia, bradycardia, epistaxis, and laryngospasm, as well as distress for the child due to discomfort from the procedure, are all recognised complications of awake flexible laryngoscopy (AFL) (2, 4). Parallel to that, the presence of secretions, floppy supraglottic structures and suboptimal views pose a challenge in making an accurate diagnosis, even in the hands of experienced paediatric otolaryngologists (5). In children younger than 3 years, non-diagnostic AFL has been reported in up to 20% of cases due to difficulty in visualising the larynx (4, 6). Furthermore, patient motion due to the uncomfortable nature of AFL may result in mucosal trauma and airway swelling. Similar complications may occur with laryngoscopy under general anaesthetic, in addition to the potential morbidity from the anaesthetic itself (1).

Laryngeal ultrasound (LUS) has emerged in recent years as a non-invasive method to evaluate vocal fold motion in children and has shown promising results. Apart from vocal fold mobility assessment, LUS has been used to diagnose laryngomalacia and vocal fold lesions in children (7, 8). While AFL is typically performed only by otolaryngologists, LUS can be performed and interpreted by radiologists, otolaryngologists, and intensive care specialists with the appropriate training (2, 9). A 2021 meta-analysis identified eight studies including 290 children with a pooled sensitivity and specificity of 91% and 97%, respectively, of LUS for identifying VFI (2). To date, there are no published studies regarding LUS in the Aotearoa New Zealand population.

Objectives

LUS has been available at our centre since 2020. The primary objective of our study is to determine the accuracy of LUS in identifying unilateral VFI in our cohort. The secondary objective of the study is to
provide an estimate of the frequency of use of LUS among clinicians around the world, as well as to elucidate
the indications, benefits, and barriers to its use, as perceived by clinicians. This is the first study to identify
VFI among children in Aotearoa New Zealand using LUS. We hypothesised that LUS is a reliable diagnostic
tool in diagnosing unilateral VFI in children.

Material and methods

LUS at Starship Children’s Health

Ethical approval

Ethical approval for this study was granted by the Auckland Health Research Ethics Committee (AH25917).

Study design + Reporting guideline

A retrospective cohort study of all patients who underwent LUS at Starship Children’s Health from 2020
to 2023 was performed. Data were obtained from electronic clinical records. There were no conflicts of
interest associated with this study. Strengthening the Reporting of Observational Studies in Epidemiology
(STROBE) guidelines were used for this manuscript.

Inclusion and exclusion criteria

To be included in the study, patients must have undergone both laryngeal ultrasound and laryngoscopy
within a period not greater than 28 days. When patients had results of both AFL and laryngobronchoscopy
under general anaesthetic (LB) available, the test performed closer in time to LUS was used for the analysis.
Individuals with non-diagnostic laryngoscopy or LUS were excluded. Those with bilateral vocal fold motion
impairment were also excluded. Reduced or absent motion in a vocal fold was considered as VFI.

Laryngoscopy

Laryngoscopy was used as the gold standard test to diagnose unilateral VFI and could be either AFL or LB.
AFL was performed by otolaryngology registrars or paediatric otolaryngology fellows or consultants with a
Storz neonatal flexible fiberscope. LB was performed under paediatric otolaryngology consultant supervision
with Storz Hopkins rod optical telescopes with or without a ventilating bronchoscope.

Laryngeal ultrasound

Transverse high-resolution ultrasonography at the level of the vocal folds and arytenoids was performed by
trained paediatric sonographers or consultant paediatric radiologists. Images were obtained with one of the
following probes: Canon linear i22LH8, Canon linear i18LX5, Philips linear L15-7io or Philips linear EL-18-4
MHz. Cine-loops were recorded for review and interpretation by consultant paediatric radiologists.

LUS survey

After analysing the data from our centre, we developed an 8-question survey exploring the use of LUS in the
paediatric population (up to 15 years of age) in other centres around the world. (Appendix 1) Google Forms
was used to create and distribute the survey. A link to the survey was sent to members of the Worldwide
Paediatric WhatsApp group (10) and The Australian and New Zealand Society of Paediatric Otorhinolaryn
gology. Attendees of the 5th Laryngology Society of Australasia Conference in Sydney, Australia were also
invited to participate via a QR code. Responses were collected during the month of November 2023.
Statistical analysis

LUS was evaluated as a diagnostic test as compared to the gold standard by calculating sensitivity, specificity, positive predictive value, negative predictive value, and Cohen’s kappa coefficient. Statistical analyses were carried out using SAS 9.4 (SAS Institute Inc., Cary, NC, USA). Statistical analysis of the survey responses was not carried out, as this was not the main objective of the study, and the sample size was considered too small to yield significant results.

Results

LUS at Starship Children’s Health

Fifty-three patients underwent LUS at Starship between July 2020 and December 2023. Twenty-three met the inclusion criteria. The age range was 7 days to 28 months (mean = 24 weeks and 3 days; median = 7 weeks and 5 days). Fourteen (61%) patients were male. Thirteen (57%) had a history of cardiothoracic surgery. Fifteen (65%) patients had unilateral VFI diagnosed on laryngoscopy, 11 (73%) of which had undergone cardiothoracic surgery. The most common indication for LUS was stridor (n=12, 52%). Other indications were weak cry (n=4, 17%), dysphonia (n=1, 4%) and feeding difficulties (n=1, 4%). In 23 patients undergoing laryngoscopy and LUS, there was agreement between investigations in 21 cases (overall concordance 91%). (Table 1) Sensitivity, specificity, positive predictive value, negative predictive value and Cohen’s kappa coefficient are presented in Table 2.

While not included in the analysis, it is worth noting that an additional 4 patients were identified to have bilateral VFI on laryngoscopy and LUS correctly identified bilateral VFI in only 1 case. Similarly, there were 3 patients suspected to have bilateral VFI on LUS, and of these cases, only 1 was confirmed on laryngoscopy. In one case, the radiologist noted bilateral vocal fold motion was present, but considered it to be limited, suspicious for paresis. However, the AFL performed on this patient 3 days later showed normal vocal fold motion.

LUS survey

Eighty-seven responses were collected. Respondents’ country of practice can be seen in Figure 1. There were seventy-two (82.8%) otolaryngologists and 15 (17.2%) speech and language therapists. Thirty-six (41.4%) respondents use LUS in their practice. Of the 36 respondents that use LUS, there were 35 otolaryngologists and 1 speech and language therapist. The most common indication for performing LUS is dysphonia/weak cry. Other common indications are stridor, as a screening test in asymptomatic children with risk of recurrent laryngeal nerve injury, and feeding difficulties, in order of frequency. Clinicians use LUS more commonly as an adjunct to laryngoscopy than as a sole investigation.

Most clinicians consider that the benefit of LUS is that it is non-invasive and safe. The main barrier identified to using LUS is lack of expertise. In the final survey question, respondents were invited to add comments in a free text field. The overarching theme was that of interest in using LUS for the detection of VFI, with non-users citing either lack of equipment or lack of expertise as their main barriers to achieving this. Users also stated that LUS had been useful in their practice primarily for identifying VFI, with indications now expanding to the diagnosis of other laryngeal pathology such as nodules, and the adoption of LUS as a point-of-care test rather than relying solely on radiologists. The indications for LUS considered by respondents were similar to ours, and, while less than half of respondents use LUS routinely, most clinicians seem to perceive it as a useful test in the paediatric population with minimal risk.

Discussion
This study demonstrates high sensitivity, positive predictive value, negative predictive value, and substantial agreement for the detection of unilateral VFI in our cohort, which is consistent with other studies (2, 11). Specificity was lower in our group than in other reports, likely due to the small number of patients with normal vocal fold function. Given its high accuracy and non-invasive nature, LUS has gained popularity for the evaluation of paediatric VFI. Although it was first described in the 1990s (12, 13), the results of our survey suggest LUS has not become common practice, although interest is increasing. As technology continues to improve and further experience is gained, LUS is gaining traction as a valuable tool for clinicians who care for children with laryngeal pathology.

It’s important to note that just over half of our patients had a history of cardiothoracic surgery. Most studies evaluating LUS for the identification of VFI have been performed solely on surgical populations with a risk of injury to the recurrent laryngeal nerve (RLN) (2). Our results suggest that LUS is translatable to non-surgical populations, but larger, prospective studies that address this are required.

Some centres are now routinely screening postoperative children with a risk of injury to the RLN with LUS (14-17). Universal screening for VFI in these children is necessary, as it has been shown that up to 47% may not present with classical symptoms of VFI (18), and LUS is a non-invasive way to do so. When combined with feeding therapist review, the accuracy of LUS may increase (17). Similarly, laryngoscopy combined with LUS is likely to clarify a diagnosis of suspected VFI compared to a single investigation. LUS has also been shown to be easy to learn for clinicians other than otolaryngologists (15, 16, 19-22), suggesting it may be a useful tool in remote areas where an otolaryngologist may not be available.

Standardised guidelines for LUS technique and for reporting of LUS are lacking. Our radiologists report LUS in a qualitative fashion, i.e., they report whether mobility is normal, reduced, or absent. Few studies have described quantitative measures such as maximum glottic angle and vocal fold-arytenoid angle to aid the interpretation of LUS (22, 23). These authors concluded that the accuracy of LUS increases by using quantitative measures. In turn, a study is currently underway to establish reference ranges for the sizes of individual structures of the larynx and trachea in the neonatal population (24).

This study has limitations. It is a retrospective review of a small sample in a single centre. The survey sample is also small and was only distributed in English. The latter, however, provides a glimpse into the frequency of use and clinicians’ perceptions of LUS, and suggests that it is a valuable, yet under-utilised resource. Laryngoscopy was performed by clinicians of varying levels of experience, and LUS was performed by several different radiologists. However, all were consultant paediatric radiologists. AFL and LB were used interchangeably as the reference test, and it’s possible that VFI was over-diagnosed in patients who underwent LB due to the effects of general anaesthetic and rigid endoscopy on the larynx (1, 25). Finally, the reference test and the LUS were not always performed within a few days of each other, with a time window of up to 28 days. However, Izadi performed a prospective study of 85 patients who had laryngoscopy and AFL within a similar window and did not show a statistically significant difference in patients whose studies were <14 days or >14 days apart (11).

**Conclusion**

LUS has demonstrated high sensitivity and specificity for detecting unilateral VFI based on our study. This is the first published study in Aotearoa New Zealand to evaluate paediatric LUS and the only survey we could identify regarding the use of LUS around the world. LUS is safe and highly accurate for the detection of VFI. Further studies are required to standardise LUS technique and interpretation. The latter will allow LUS to be more widely used, including as a point-of-care investigation, by both otolaryngologists and other clinicians.

**Other information**

**Funding:** There was no funding associated with this work.

**Appendices**
Appendix 1. Survey

Paediatric Laryngeal Ultrasound

We would be grateful if you completed this survey exploring the use of laryngeal ultrasound (LUS) in the paediatric population (up to 15 years of age)

What country do you practice in?

What is your clinical role?

Otolaryngology - Head and Neck Surgeon (OHNS)

Doctor - non OHNS

Speech and Language Therapist

Do you use LUS in your practice?

Yes

No

In your practice, what are the indications for performing LUS? Please select all that apply.

Asymptomatic children, i.e. as a routine postoperative screening test where there is risk of recurrent laryngeal nerve injury.

Stridor

Dysphonia/weak cry

Feeding difficulties

Other

Do you use LUS as . . .

A sole investigation

An adjunct to laryngoscopy (either awake flexible laryngoscopy or laryngoscopy under general anaesthetic)

I don’t use LUS.

In your practice, what are the barriers to using LUS?

Cost

Lack of staff trained in LUS/lack of expertise.

Lack of equipment

In your view, what are the benefits of using LUS?

Cost

More accessible than ORL services

Non-invasive

Safe in high-risk patients

If you wish to make any further comments regarding paediatric LUS, please write them here.

Tables
Table 1. Results of LUS and laryngoscopy

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<th>Mobile vocal folds</th>
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Table 2. Accuracy of LUS

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<th>Positive predictive value (95% CI)</th>
<th>Negative predictive value (95% CI)</th>
<th>Cohen’s kappa coefficient (95% CI)</th>
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Figures

Figure 1. Survey respondents’ country of practice.
References


