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Case Report
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# The Challenges of Diagnosing Paradoxical Vocal Fold Movement in Infants and the Potential Role for Bedside Ultrasound: A Case Report

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## Abstract

**Objectives:** This manuscript reviews some of the practical difficulties in diagnosing abnormalities of vocal fold motion in neonates, with the potential use of ultrasound to address some of these challenges.

**Methods and Results:** We present a case report of a 6-week-old infant, referred following cardiac surgery for investigation of stridor exacerbated during breastfeeding. We assessed vocal fold mobility and swallow utilizing using Flexible Endoscopic Evaluation of Swallowing (FEES) and laryngeal ultrasound. Both were performed at bedside during breastfeeding, with Paradoxical Vocal Fold Movement (PVFM) confirmed by both modalities.

**Conclusion:** This case report illustrates the potential utility of bedside ultrasound in diagnosing Vocal Fold Movement Impairment (VFMI) in infants. Ultrasound is a portable, low risk, noninvasive imaging modality that appears to complement the clinical findings on flexible endoscopy in this cohort, whilst overcoming some of the challenges in diagnosing VFMI in infants.

**Keywords: Ultrasound; Larynx; Vocal fold; Infant; Breastfeeding; FEES; Paradoxical**

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## Introduction

Vocal Fold Motion Impairment (VFMI) is a term that encompasses a range of disorders of vocal fold movement including immobility of one or both folds and impaired coordination of movement, as seen in Paradoxical Vocal Fold Movement (PVFM). In infants, the spectrum of VFMI disorders can present with similar symptoms, including a weak cry, stridor, feeding difficulties and possible aspiration [1,2]. There is also overlap in presenting symptoms with a range of other airway anomalies, such as laryngomalacia and less common congenital structural anomalies of the larynx and subglottis.

Paradoxical Vocal Fold Mobility (PVFM) is a subcategory of VFMI, where intermittent episodes of stridor are caused by active adduction of the vocal folds on inspiration. It is a well-recognized diagnosis in adolescent and adult patients [3], but the diagnosis can be challenging as it usually requires viewing of vocal fold movement during episodes of abnormal movement. This is particularly difficult as episodes of abnormal vocal fold movement are usually intermittent and can be triggered during specific activities where concurrent endoscopy is challenging. Ultrasound has been reported as aiding in diagnosis of PVFM in a case report of a teenage athlete [4].

Literature published on PVFM in infants is sparse, generally reporting isolated cases or small series [5,6]. Visualization of an infant's vocal folds is challenged by the need for specialized equipment and expertise. Variable tolerance of the procedure and supraglottic soft tissues also often limits a direct view of the vocal folds. As PVFM symptoms are usually intermittent, diagnosis can be further challenged by the need to visualize the vocal fold movement at the time stridor is generated. As such, PVCM is not a well-recognized differential diagnosis in infants who present with intermittent stridor [6].

The presenting symptoms of PVFM, unilateral vocal fold immobility and laryngomalacia can

be similar. However, the ideal clinical management is quite different. Therefore, an accurate diagnosis and differentiation between these conditions has clinical importance. In all these conditions, inspiratory stridor may be exacerbated or only present during crying and feeding, and as such, visualization during feeding may be necessary to diagnose the cause of stridor.

It is also recognized that infants with VFMI have a high incidence of silent aspiration, meaning that an instrumental assessment of swallowing may be warranted, as the absence of overt signs of aspiration does not exclude the possibility that aspiration is occurring in this cohort [1,7,8].

Flexible trans-nasal laryngoscopy is considered the gold standard in diagnosis of vocal fold movement impairment in young infants [9]. However, the procedure requires specialized equipment, significant technical expertise and is considered to have only moderate reliability [10]. It is recognized that recording and then reviewing endoscopic images improved accuracy of diagnosis [11]. However, even careful review of recorded imaging can be compromised by poor patient tolerance of the procedure, the presence of secretions, retroflexed epiglottic positioning and the dynamic prolapse of supraglottic soft tissues that impairs visualization of the vocal folds in infants with laryngomalacia.

In 1997, the alternative of using ultrasound to assess vocal fold motion in infants and children was proposed as a non-irradiating and non-invasive investigation [12]. However, the potential clinical utility of ultrasound has really only recently been promoted, with an increasing number of publications over the last 5 to 10 years [13]. Ultrasound image quality and definition has improved over the intervening two decades. Technological advances now enable imaging of vocal fold structure, even to the level of detail that allows assessment of the relative concentration of elastin and collagen fibers [14]. Laryngeal ultrasound is now successfully used for a broader range of diagnostic applications, including the diagnosis of vocal fold nodules and laryngomalacia [15,16].

Using laryngeal ultrasound to assess vocal fold mobility in adults has remained limited, however, hindered by calcification of the thyroid cartilage and intraluminal air. But the sonographic appearance of the infant larynx, which is usually not yet calcified, allows capture of high-quality definition of anatomical structures and movement [17]. Accordingly, ultrasound has been reported as achieving conclusive images for analysis of vocal fold movement in only 35% of adults [18], but in up to 92% of infants [9].

## Case Presentation

Written consent was given by the infant's family for presentation as a case report and use of images. We present a male term infant, born *via* a normal vaginal delivery with a birth weight of 3012 g. Respiratory compromise was present at birth, requiring endotracheal intubation, performed without technical difficulty at 3 h post-delivery. An aortic coarctation with a large patent ductus arteriosus was diagnosed, with an open repair via sternotomy performed at 6 days of age. His postoperative course was complicated by initial left lung collapse with respiratory distress and a prolonged requirement for continuous positive airway pressure support. He was weaned gradually to daytime high-flow oxygen over a period of weeks. A formal airway endoscopy under general anesthetic at 5 weeks of age identified laryngomalacia (Figure 1) with no other anatomical anomalies of the airway.

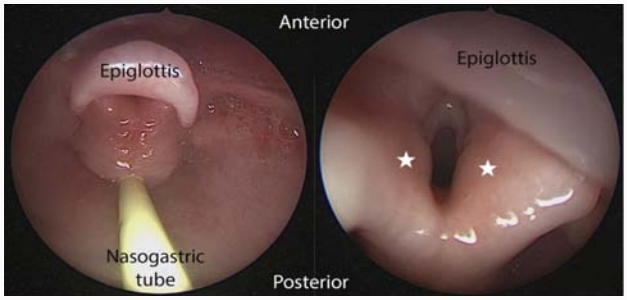


**Figure 1:** Images of supraglottis and larynx captured during rigid endoscopy under general anesthetic.
White stars: Arytenoid cartilages with overlying mucosa.

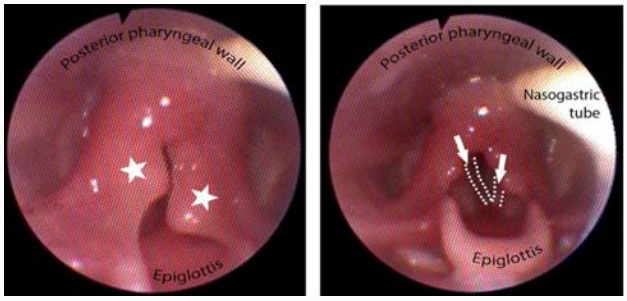


**Figure 2:** Flexible endoscopic images of supraglottis and larynx.
White stars: Aryepiglottic folds.
White arrows: Vocal folds.

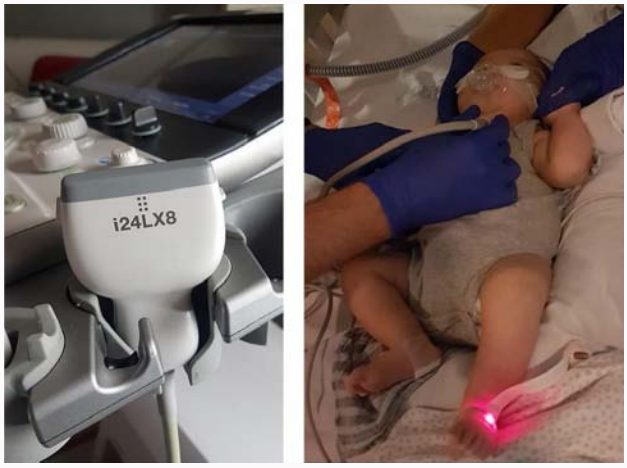


**Figure 3:** Bedside capture of vocal fold mobility using ultrasound.

Nutrition was initially intravenous, with gradual transition to nasogastric tube feeding. The infant's weight gain remained stable on the 9th percentile. Attempts at feeding at the breast started at around 4 weeks of age, with respiratory effort and noise significantly exacerbated. Consistent audible secretions moving in the airway raised concern about possible aspiration. A multidisciplinary feeding assessment was undertaken by the pediatric speech-language pathology and otolaryngology teams. A bedside Flexible Endoscopic Evaluation of Swallowing (FEES) was followed by a bedside laryngeal ultrasound, performed by a pediatric radiologist. Both modalities included assessment during breastfeeding.

FEES confirmed the presence of laryngomalacia, with dynamic prolapse of supraglottic soft tissues over the laryngeal inlet on inspiration (Figure 2). The endoscopic view of the vocal folds was compromised by dynamic soft tissue prolapse on inspiration, but

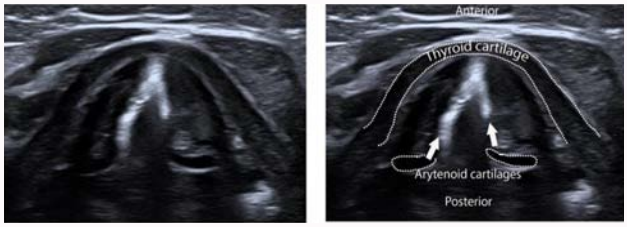


**Figure 4:** Ultrasound images of larynx and vocal folds (white arrows).

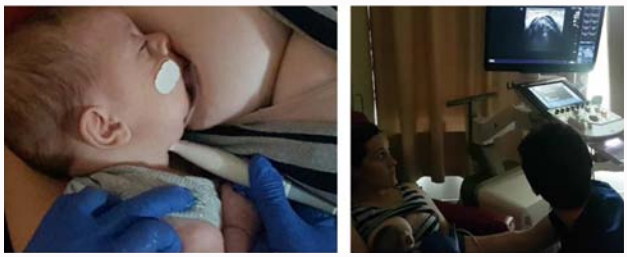

**Figure 5:** Capture of vocal fold movement during breastfeeding using ultrasound.

complete obstruction of the laryngeal inlet did not occur. There was no delayed swallow, aspiration events or post swallow residue visualized during the FEES. Slow motion review of the recorded endoscopy (with audio) confirmed the diagnosis of PVFM, capturing sequences of normal vocal fold motion as well as intermittent episodes of vocal fold adduction on inspiration that coincided directly with audible stridor. The PVFM was exacerbated by breastfeeding in this infant so an awake transnasal flexible assessment of his airway would likely have missed the diagnosis as it would not capture events occurring during feeding.

The following day, a bedside ultrasound was performed by a consultant radiologist using a Canon Aplio i800 ultrasound machine with i24LX8 and i22LH8 (hockey-stick) probes (Figure 3). The infant's head was gently stabilized by an assistant and a pacifier helped keep him settled at initiation of the procedure. Image quality was sufficient to enable assessment of laryngeal anatomy including identification of the vocal folds (Figure 4). Dynamic imaging captured normal vocal fold movement at rest, with intermittent, brief episodes of vocal fold adduction during inspiration with temporal correlation with the presence of stridor. Dynamic imaging during breastfeeding (Figure 5) captured sustained episodes of bilateral vocal fold adduction with associated exacerbation of respiratory effort and increased volume of stridor.

## Discussion

This case study demonstrates the potential for ultrasound as an effective and practical diagnostic tool for assessing vocal fold mobility in infants. We agree with Zhang et al., that ultrasound may provide supplementary information to that provided by flexible laryngoscopy [19], particularly when a clear endoscopic view of the vocal folds cannot be achieved. Once an endoscopic diagnosis of any form of VFMI has been made, ultrasound appears also to be an ideal modality for monitoring vocal fold mobility over time, as success of trans-nasal flexible endoscopic assessment can be limited by patient tolerance in young children [20].

The potential for the ultrasound to be done at bedside brings added benefits in timeliness, convenience and safety. This can minimize the risks associated with moving a patient out of a monitored environment, which is particularly desirable with post-operative cardiac patients, as in this case report. For more stable patients, there is potential for the ultrasound to be done in the otolaryngology outpatient clinic setting. Ultrasound has become a tool used at point of care for a range of indications by specialists outside of the radiology unit, including use in emergency rooms and surgical outpatient clinics [21-23]. Although formal diagnostic ultrasound is best performed by formally trained sonographers, the use of ultrasound by surgeons for specific screening or indications such as assessment of vocal fold movement is becoming increasingly accepted. A study assessing otolaryngologist performance of laryngeal ultrasound has shown a relatively rapid learning curve for acquisition of appropriate skills [24].

However, further reporting of the clinical utility of ultrasound in clinical practice is needed to determine the strengths and limitations of ultrasound of the larynx, particularly in the context of airway and swallowing dysfunction in infants and young children.

## Conclusion

We believe that disorders of VFMI are possibly underdiagnosed due to challenges in viewing vocal fold movement in infants. We believe an early and accurate assessment of vocal fold movement and screening for the presence of aspiration is warranted in infants presenting with airway and/or feeding dysfunction. This is particularly important in cohorts recognized to have increased risk of VFMI, such as infants following aortic arch surgery [25]. We consider ultrasound to be a suitable non-invasive modality for assessing vocal fold movement, when flexible endoscopy is not readily available or an adequate endoscopic view of the vocal folds is not possible. We encourage ultrasound to be considered as a suitable tool to screen for VFMI following any surgery associated with risk of damage to the recurrent laryngeal nerve, as well as for monitoring the return of function over time when a diagnosis of VFMI has been made.

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